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THE SEMON LECTURE, 1960

CLINICAL ASPECTS OF CONGENITAL ANOMALIES OF THE LARYNX, TRACHEA, BRONCHI AND ŒSOPHAGUS

PAUL H. HOLINGER, M.D.*, Chicago, Illinois, U.S.A.

THE great man whom we honour here, one of the founders of our specialty, would accept, I am sure, today's broad aspects of the field he did so much to establish. In a day when specialization was far from routine, Sir Felix Semon understood the need of concentration of effort in one phase of the expanding intricacies of medicine and surgery. His background as a physician was a basic prerequisite to specialization and it is essential for the best interest of patient and physician alike that this pattern be maintained.

The manner in which we receive the experience and knowledge of our predecessors, probably because the mass of factual information is overwhelming, usually gives us little opportunity to reflect on the effort they made and the amount of work they did in gaining an understanding of each new point. The honour you have extended to me in asking me to give the Semon Lecture, gave me an opportunity to review the life and work of Sir Felix Semon. I am indebted to Scott Stevenson (1949) for his resumé of the history of Laryngology and to the interesting Semon Lecture of 1957 of Mr. F. C. W. Capps (1958) in which he reviewed the

* From the Department of Broncho-œsophagology, The Children's Memorial Hospital, and the Departments of Otolaryngology, Presbyterian-St. Luke's Hospital, and the University of Illinois, College of Medicine.

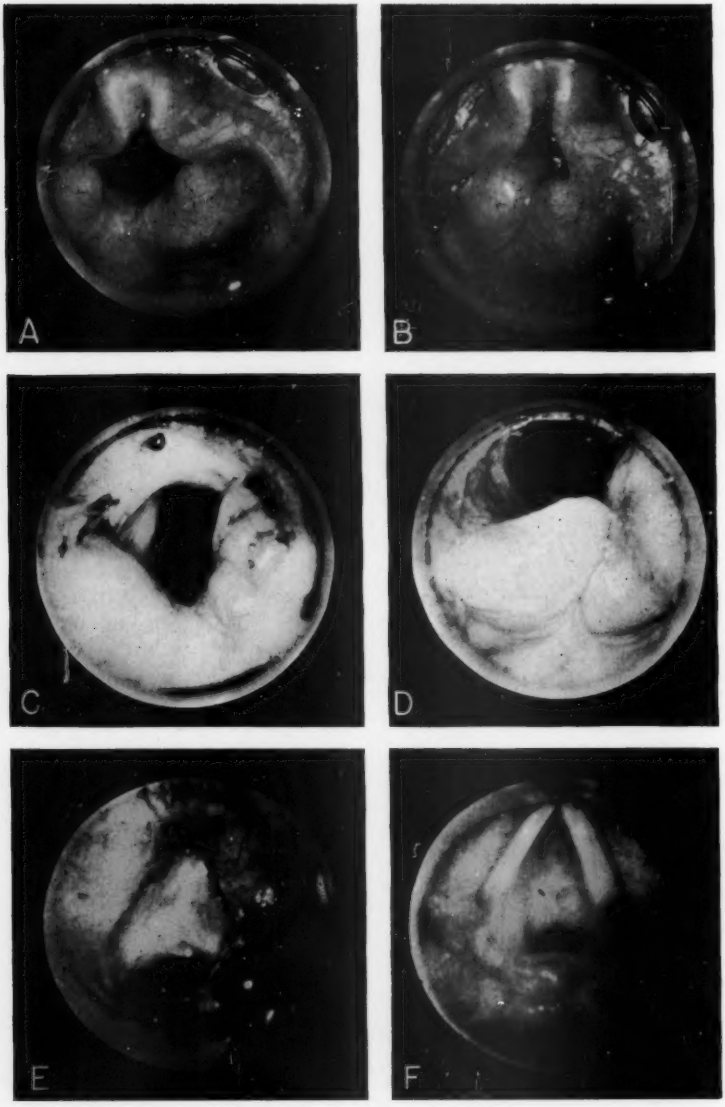

FIG. 1.

Direct laryngoscopic photographs of the larynx showing congenital laryngeal anomalies
A and B.—Expiration and inspiration, respectively of an infant three months of age with congenital stridor.
C.—Congenital supraglottic web between the aryepiglottic folds at the level of the false cords.

work so indelibly associated with the man we honour here; this work, summarized in a statement, still controversial, but known to every student and examiner as Semon's Law. I am indebted, too, to Mr. Myles Formby for sending me numerous references concerning Sir Felix Semon (McBride, P., 1913) and to Sir Douglas Logan for the list of distinguished previous Semon Lecturers with the titles of their discussions. This, truly, is a history of our specialty and gives me an even greater sense of responsibility as well as pride in accepting this honour.

The subject I have chosen, Congenital Anomalies of the Larynx, Trachea, Bronchi and Œsophagus, is obviously too broad to cover in its entirety. The most complete text of Sir StClair Thomson, Sir Victor Negus and Mr. Geoffrey Bateman serves as a constant reference with its excellent descriptions of even the rarest entities (1955). Furthermore, because of limits of time and space, we must forego a discussion of the important and interesting embryologic development of these anatomical areas. The clinical aspects themselves can be but briefly reviewed and will be presented from the basis of personal experience. In this way, we hope to stress the rôle of endoscopy in otolaryngology and the opportunity as well as the responsibility of the otolaryngologist in endoscopy (Holinger *et al.*, 1960). The otolaryngologist, trained in monocular surgical technics is prepared as is no other specialist to perform the endoscopic diagnostic and therapeutic procedures that are essential in the management of respiratory and œsophageal problems in the newborn. Pædiatric laryngology itself has many new ramifications. In addition, the work of the pædiatric surgeon has opened an area in this field which is challenging, and rewarding. The laryngeal, tracheal, bronchial and œsophageal phases of modern pædiatric surgery demand a co-operation between specialties to solve these problems of mutual interest.

The Larynx

Congenital Laryngeal Stridor. So-called congenital laryngeal stridor is a transient form of stridor in infants which is associated with an abnormal flaccidity of the laryngeal tissues (Fig. 1). "Laryngomalacia" is a more satisfactory term since stridor is a symptom of respiratory obstruction rather than a specific disease entity (Kelemen, 1953). However, in infants with stridorous, noisy respiration, a differentiation must be made as to ætiology, and the majority of congenital laryngeal anomalies

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D.—Flaccid, elongated soft parts of the left arytenoid in an adult being drawn into the larynx during inspiration. The right arytenoid followed over the left as inspiration progressed.

E.—Congenital fusion of the anterior half of the cords simulating a laryngeal web.

F.—Symmetrical subglottic stenosis at the level of the cricoid cartilage.

(Figs. 1E and F are enlargements of frames of 16 mm. motion picture films.)

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recorded in this discussion were referred by pædiatricians with the admitting diagnosis of congenital stridor. Laryngomalacia did constitute the largest number of such referrals, but a description of this entity is hardly necessary here. A number of reviews of the condition appear in texts (Lederer, 1952; Jackson and Jackson, 1959). Motion pictures of the infant larynx taken through the direct laryngoscope illustrating this discussion show the tubular epiglottis and fluttering arytenoids that produce the characteristic stuttering component of the cry. These movements, so much slower than the phonatory vibrations of the newborn discussed by Professor Rüedi here two years ago (1959), are still, obviously, passive.

TABLE I.

CONGENITAL WEBS OF THE LARYNX.

Supraglottic	—	4
Glottic	—	19
Membranous web	15	—
Fusion of anterior half of cords	2	—
Transparent membrane	1	—
Total glottic atresia	1	—
Subglottic	—	2
Total		25

A number of infants with congenital stridor are noted to have a funnel breast (*pectus excavatum*). This usually disappears as the stridor subsides at 1½ to 2 years of age. In some cases, of course, it is permanent and requires surgical correction; but the association between laryngeal obstruction and *pectus excavatus* is important.

Congenital stridor is rarely severe enough to require surgical intervention. However, in two extreme cases tracheotomy was necessary because the infants laboured so hard at breathing that they were cyanotic and totally exhausted with feedings. Following tracheotomy, weight gain was rapid.

Congenital Subglottic Stenosis. This clinical picture is a difficult one to evaluate. Infants falling into this category constituted the largest single group of congenital laryngeal anomalies requiring tracheotomy under one year of age (Holinger *et al.*, 1954). Furthermore, it is apparent that this condition more than any other congenital lesion is responsible for the prolonged need of the tracheotomy, some requiring the tube until two or three years of age. The condition consists of thickening of subglottic

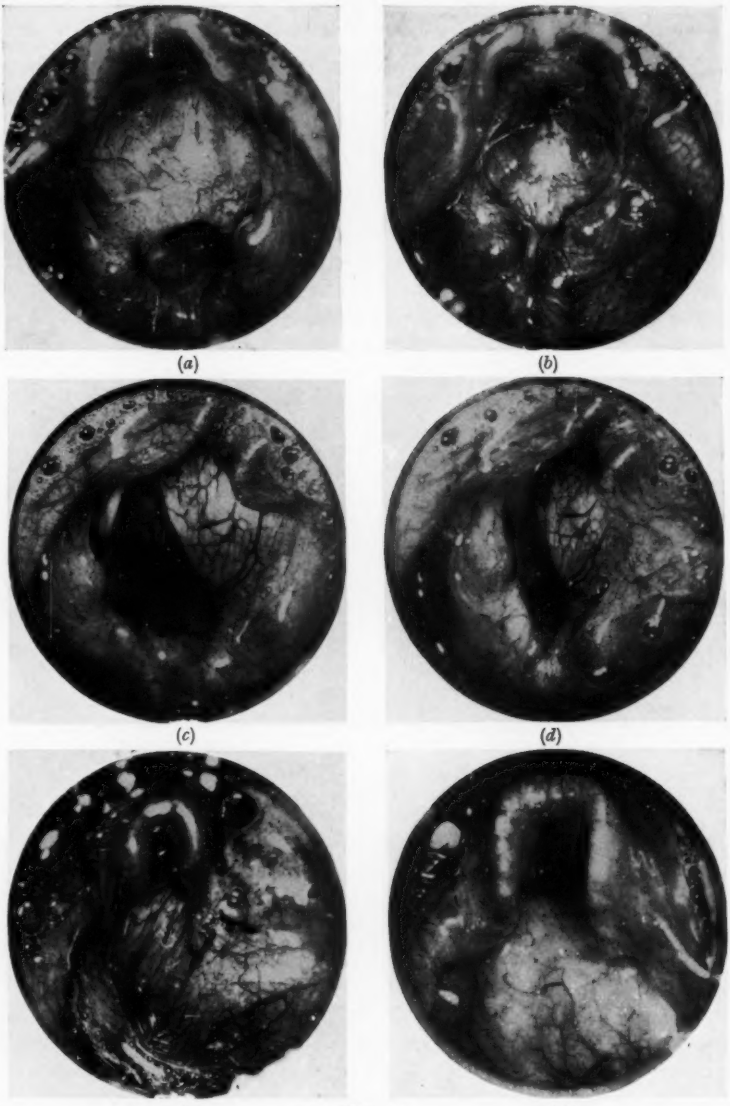


FIG. 2.

Direct laryngoscopic photographs of the larynx showing congenital laryngeal anomalies.

A. Inspiration—B. Phonation of a ten-day-old infant with a supraglottic web.

C. Inspiration and D.—Phonation of a three-week-old infant with severe stridor due to a laryngocele seen protruding from the right laryngeal ventricle.

E.—Laryngocele involving the right aryepiglottic fold and arytenoid of a newborn infant. An emergency tracheotomy was done a few hours after birth.

F.—Similar laryngocele in a three-year-old child whose X-rays are shown in Fig. 6A and B.

structures and sometimes of the true cords themselves to cause respiratory obstruction of a severe degree in newborn infants. The least laryngeal inflammation precipitates a tracheotomy since the limiting cricoid cartilage does not permit swelling of tissues in any direction other than inwardly, at the expense of the airway. The point of greatest obstruction is generally 2-3 mm. below the level of the superior surfaces of the true cords. Biopsy of one such area showed no inflammatory reaction. The earliest symptom of this condition beginning at birth was stridor, with or without episodes of cyanosis depending upon the severity of the stenosis. In mild cases, frequently recurring episodes of "croup" were the only suggestion of this condition. Every respiratory infection simulated

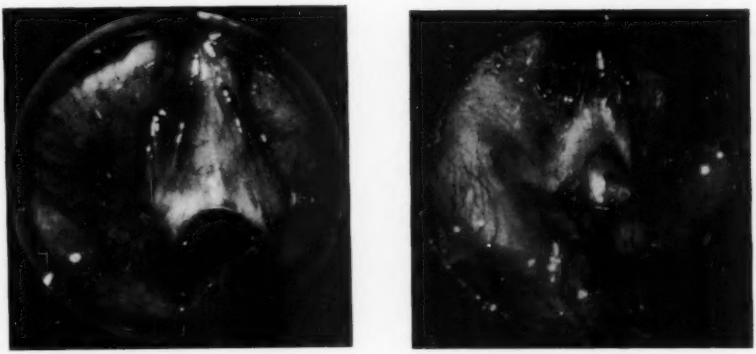
FIG. 3.
Web involving the anterior commissure of a 31-year-old female who had been hoarse since birth.

laryngotracheobronchitis and although the episodes were characterized by severe obstruction, there was minimal evidence of inflammation.

Congenital Webs. Congenital webs of the larynx most frequently consist of membranes across the anterior half or two-thirds of the vocal cords. However, supraglottic and subglottic webs should be included in this category. Twenty-five patients with laryngeal webs constitute this group of anomalies (Table 1). In nineteen the webs were glottic, in four supraglottic and in two, subglottic. Three of the supraglottic webs were fusions of the false cords above an apparently normal glottis (Figs. 2*a* and *b*). In the fourth, the fusion was almost cartilaginous in consistency and the infant had a fusion of the anterior half of the true cords as well.

In nineteen the webs were glottic and these were further divided into four types; fifteen were definite membranes between the cords consisting of a web across the anterior portion of the glottis at the level of the superior surfaces of the cords, through which the outlines of the phonating edges of the cords could be distinguished. The degree to which the glottis

was covered varied from small anterior commissure webbing (Fig. 3), to almost complete closure of the glottis (Fig. 4*a*). In the majority of cases the anterior two-thirds of the glottis was involved, the tough membrane originating along the entire cord with the exception of the vocal process of the arytenoids; it arched anteriorly to give a posterior chink for respiration. In two infants the webs consisted of a fusion between the anterior half of the cords, quite unlike a membranous web (Fig. 1*e*). The anterior half of the phonating edges of the cords appeared to be in direct apposition without an intervening membrane and the cords separated posteriorly in an inverted "V" rather than in a "U" shape characteristic of the glottic chink in the more commonly encountered web. One newborn infant had a



(*a*) FIG. 4. (*b*)

Direct laryngoscopic photographs of
A.—Congenital web of the larynx of a 7-year-old girl.
B.—Congenital atresia of the larynx.

thin, transparent membrane across the superior surface of the cords which was seen when it was examined with a direct laryngoscope because it made no sound as it cried. The membrane was split with laryngeal forceps and the edges retracted immediately and the appearance of the larynx, the cry, and respirations became entirely normal.

The nineteenth infant with a glottic web had a total membranous atresia of the larynx (Fig. 4*b*). A tracheotomy was performed at birth because of failure of pulmonary aeration in spite of strong, active respiratory efforts. Subsequent laryngoscopy demonstrated the atresia shown in Fig. 4*b*. Congenital subglottic webs are described as relatively common and probably constitute the condition herein referred to as subglottic stenosis. In this classification we have included only two infants with web-like subglottic obstruction (Fig. 1*f*). Both proved to have anomalies of the cricoid cartilage. Clinically they appeared as webs at the level of the cricoid with a small opening posteriorly. The walls were cartilaginous in

their rigidity and attempts to dilate the lumen were unsuccessful, necessitating tracheotomy during the first hour of life (Holinger *et al.*, 1954).

Congenital Cysts and Laryngoceles. Most congenital laryngeal cysts described in the literature are confined to the immediate neighbourhood of the ventricular appendix. It is probable that cysts are the result of simple disturbance in fœtal development and not genuine embryonic malformations. Congenital cysts and laryngoceles have a similar development, but cysts do not communicate with the interior of the larynx. Laryngoceles admittedly become clinically perceptible only when swollen by air forced into them, or when filled with a collection of fluids; thus it is

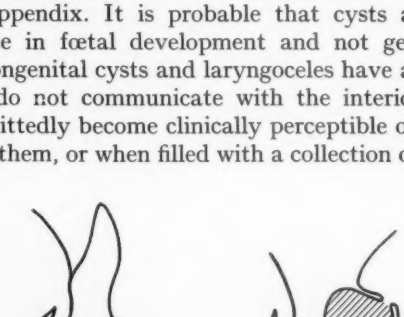


FIG. 5.
Diagrammatic representation of site of two types of laryngoceles observed in infants.

obvious that laryngoceles and cysts are often indistinguishable from each other. In contrast to laryngeal webs, congenital cysts rarely are seen first in adults, probably because as hollow structures they fill and grow in intra-uterine life and are, therefore, observed directly after birth. Laryngoceles, however, may manifest themselves at any age. In newborn infants they produce alarming symptoms of respiratory obstruction and aphonia. Lateral X-rays of the neck suggest the nature of the lesion and confirmation is obtained by direct laryngoscopy.

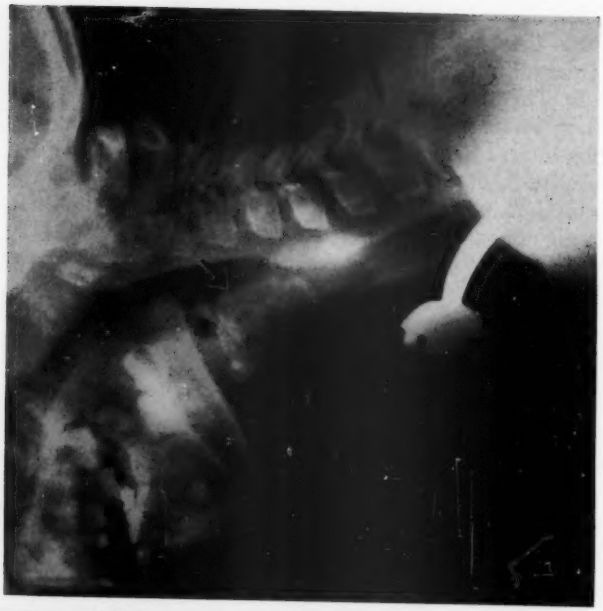
Of a total of 29 patients observed who have had proven laryngoceles, eight were infants under two months of age and one a child three years of age who had had laryngeal obstruction since birth. All others were adults. Five of the nine required tracheotomies, three during the first 24 hours after birth. The laryngoceles were of two types. One appears to have dissected posteriorly from its origin in the Sinus of Morgagni into the aryepiglottic fold and arytenoid (Figs. 6*a* and *b*). It presents itself as a round, cystic mass, quite tense and somewhat translucent. The contour of the hemilarynx involved is completely lost and the tiny epiglottis seems

perched on top of the cyst. Reflecting it aside, the opposite side of the larynx has been found to be normal except for compression and lateral displacement. Four infants with this anomaly were seen at birth or shortly thereafter. Therapy consisted of laryngoscopic aspiration of the contents of the laryngocele with a laryngeal syringe and needle to be repeated when obstructive symptoms became apparent. Three of the four infants required a tracheotomy. Aspiration may be necessary two or three times a year in infancy, less frequently later. The oldest patient, now fifteen years of age, was less than a week of age when first seen. Her airway was maintained in infancy without a tracheotomy by occasional aspiration of the cyst. She has required aspiration only twice in the past five years. Resection through an external pharyngotomy or laryngotomy hardly seems warranted.

The second type of laryngocele observed appears as an eventration of the ventricle. The direct laryngoscopic photograph depicts the condition and the diagram suggests the probable manner in which it is formed (Fig. 5). Five infants with this anomaly have been observed, four under two months of age and one three years of age (Fig. 6*c*). The symptoms were dyspnœa, stridor and aphonia. Visualization of the transparent, cystic mass protruding from the ventricle established the diagnosis. Therapy consisted of incision of the cyst to release its contents of gelatinous mucus and removal of the frayed edges of the redundant mucosa with laryngeal cup forceps. Two of the infants required a temporary tracheotomy.

The thyroglossal duct cyst should be mentioned at this point since it must be considered in the differential diagnosis of acute laryngeal obstruction in the newborn. Its position at the base of tongue causes the epiglottis to be pushed back, trap-door fashion, over the larynx. Needle aspiration immediately relieves the obstruction.

Hemangiomas. The association of laryngeal obstruction with lymphangiomas and hemangiomas of the head and neck presents a serious problem of diagnosis and treatment (Baker *et al.*, 1956). Direct involvement of the larynx by the process is shown in the illustrations of X-rays (Fig. 6*d*) and the extent of two such lesions is recorded in motion pictures. When infants with these processes require a tracheotomy during the first few days of life the prognosis must be guarded. The condition appears to increase in severity until the six month age is reached, when some spontaneous regression has been noted. Four adults with cavernous hemangiomas have been seen, one of whom an 18-year-old female required an extensive neck dissection because of a gradual increase in the size of the vascular pattern in the larynx. Involvement of the cervical vertebrae was apparent on radiologic examination of the neck. Seven infants with laryngeal hemangiomas have been observed whose ages ranged from a few days to ten months. All required tracheotomies. Various forms of radiation have been employed in some with a full realization of its effect



(b)

(a)

(c) (d)

FIG. 6.
Lateral X-rays of the neck of infants with congenital laryngeal anomalies.
A.—Laryngocele extending into the aryepiglottic fold.
B.—Air injected into the sac after aspiration of fluid contents.
C.—Laryngocele bulging into ventricle.
D.—Lymphangioma involving the epiglottis of an infant.

on the growth of the larynx, but with no alternative when spontaneous regression failed to materialize.

Laryngeal Paralysis. The paralysis of one or both vocal cords in infants is often a serious laryngeal manifestation of a gross anomaly of another organ system. In his review of abductor paralysis, Capps (1958) referred to large series to analyse ætiologic factors. All seem to deal with paralysis in adults, but one sentence alludes to this significant condition in infants; namely, in operating on a patent ductus, damage to the left recurrent laryngeal nerve may cause respiratory obstruction, which with even minimal post-endotracheal anæsthesia œdema could require a tracheotomy. In our experience, central nervous system anomalies as well as congenital anomalies of the heart, great vessels, œsophagus or tracheobronchial tree may be complicated by laryngeal paralysis. Pre-operatively, a large congenital heart, double aortic arch, or bronchogenic cyst have been found as ætiologic factors of paralysis of the left cord in the newborn. Left cord paralysis has been noted post-operatively following the correction of these conditions as well as following some of the extensive surgical reconstructions for œsophageal atresia and fistulae.

Dr. Florence Cavanagh of Manchester, England, discussed the subject in a paper entitled "Vocal Palsies in Children" (1955). In 107 laryngoscopies in infants and children, 37 were found to have cord paralysis. Eleven had symptoms at birth, five developed symptoms a few days after birth and the remainder showed signs of paralysis from a few weeks to seven years of age. Ten infants had paralysis of both cords, four were left cord paralysis and 23 had right cord paralysis. In succeeding examinations, partial recovery was noted in some in a few months and complete recovery came after as long a time as 2½ years. Only two cases showed no sign of recovery. The chief ætiologic factors were considered to be birth injury and disease of the mother during pregnancy. Infectious diseases, measles, pneumonia and tuberculosis were mentioned as presumed causes. It is somewhat difficult to understand the relatively high incidence of right vocal cord paralysis. However, since a general anæsthetic had been administered to permit the examination, it is possible that vocal cord function was altered by the anæsthetic, or by the direction of the laryngoscope.

During a thirteen-year period, 1947 to 1959 inclusive, we have observed 63 infants under 18 months of age with unilateral or bilateral vocal cord paralysis; 34 were male, 29 female (Table II). In 42 infants the paralysis was bilateral, either partial or total. 21 of these were male, 21 female; eight infants, five male and three female, had a unilateral right vocal cord paralysis while thirteen infants, eight male and five females, had a unilateral left vocal cord paralysis. The diagnosis in each instance was established through the examination of the larynx by direct laryngoscopy without anæsthesia.

Bilateral Recurrent Laryngeal Nerve Paralysis. In 17 of the 42 infants with bilateral paralysis, the paralysis was complete with both cords lying in a midline position without abduction of any degree on inspiration. Obviously respiratory obstruction was severe and usually acute. In 25 infants the bilateral paralysis was considered partial because very minimal activity (abduction) of one or both cords could be observed during a prolonged direct laryngoscopy.

Bilateral Complete Paralysis. Of the 17 patients in this group, 11 are living, five are known dead and the status of one is unknown, although presumed dead. Thirteen were tracheotomized, including the one whose

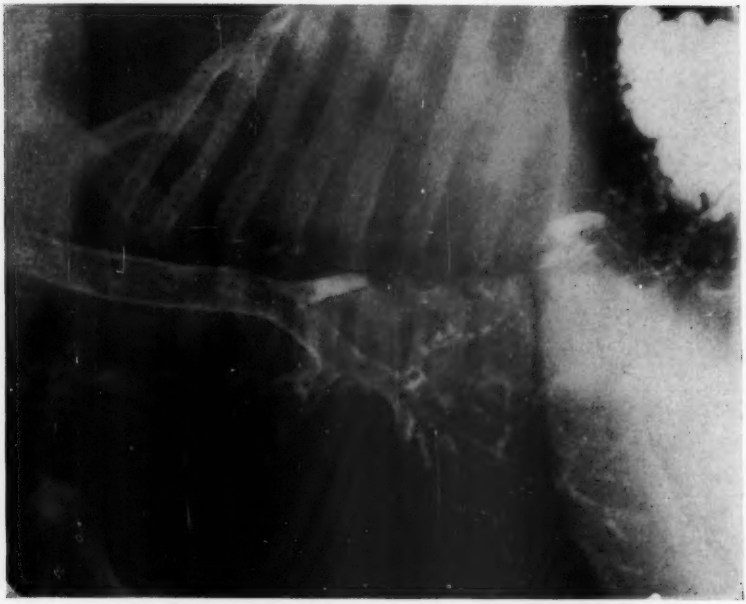
TABLE II.

LARYNGEAL PARALYSIS IN INFANTS

		Tracheotomized	Central nervous system lesions	Congenital heart disease	Died	Living with tube	Extubated
Bilateral—42							
Complete	17	13	9	0	5	5	3
Partial	25	7	13	2	0	7	0
Unilateral—21							
Right	8	1	3	0	0	0	1
Left	13	1	1	9	1	0	0
Total	63	22	26	11	6	12	4

status is unknown, and of these, eight are living; five of the eight are still tracheotomized. Of the eight patients tracheotomized who are still living, three were apparently normal at birth except for the laryngeal paralysis; two of the thirteen had to be tracheotomized within the first 24 hours of life, most of the others during the first month, and one at eight months of age. Nine of the seventeen infants had evidence of other congenital neurological lesions; four had myeloceles or meningoceles, two had marked mental retardation, two had severe cerebral birth injuries, and one a hydrocephalus. Two of the seventeen infants had acquired neurological lesions responsible for the bilateral laryngeal paralysis; one from whooping cough encephalitis, one from a button in the œsophagus for nine months with cervical mediastinitis and scar involving both recurrent laryngeal nerves. Two of the seventeen infants had an associated congenital subglottic stenosis and the remaining four were apparently normal at birth except for the paralysis.

Bilateral Partial Paralysis. There are 25 infants in this group. Symptoms were aphonia or a weak breathy cry, stridor and varying degrees of dyspnœa. The infants were considered to have a bilateral



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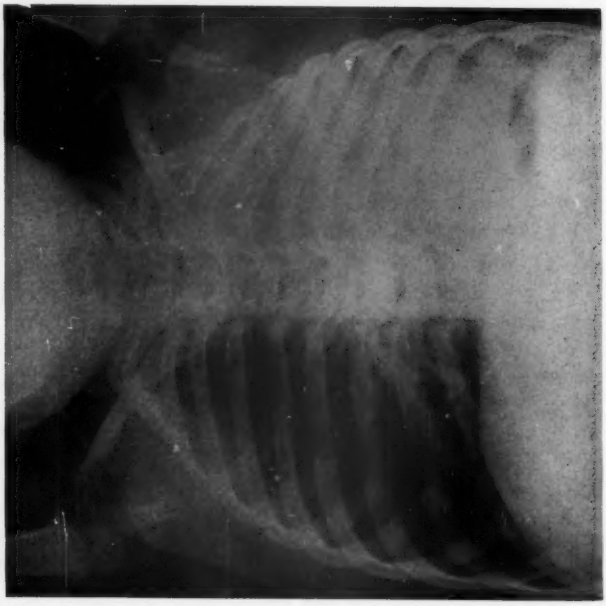

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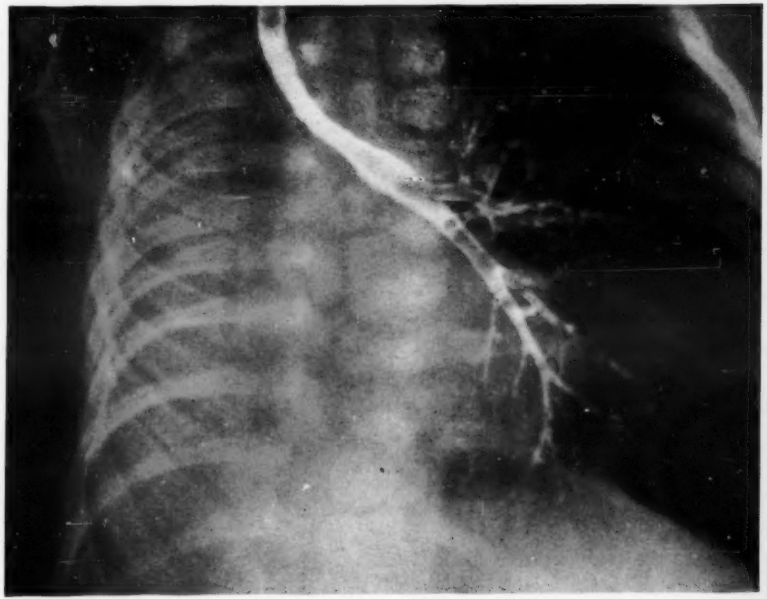
(a) (b)



(c) (d)

FIG. 7.

A and B.—Agenesis of the left lung. The bronchogram shows a small bronchial stump.
C. and D.—Agenesis of the right lung. The bronchogram shows a total absence of the right bronchus.

partial paralysis of the larynx when direct laryngoscopy demonstrated a flaccid larynx with 25 per cent. or less abduction of both cords, or when one cord was completely flaccid and motionless in the midline and the opposite cord limited to less than 25 per cent. of its motility. Fourteen infants (eight male, six female) were considered to have such a partial bilateral paralysis. Three required tracheotomy. Nine had other neurological lesions such as pharyngeal paralysis, mental retardation or other evidence of birth injuries while in one, the paralysis was associated with an encephalitis acquired at one month of age. One infant had a transposition of the great vessels, and one a diaphragmatic hernia; this infant died following surgical repair of the hernia. The remaining three infants were apparently normal except for their laryngeal dysfunction. Four of the fourteen infants died of conditions associated with their central nervous system disorders, of which the laryngeal paralysis was a part. One infant died at three years of age, more than two years after examination, of bulbar poliomyelitis. Thus, in partial as well as in total bilateral laryngeal paralysis, the laryngeal lesion is usually but one manifestation of a generalized neurological disorder.

Seven infants (four male, three female), were observed who had a total right and a partial left cord paralysis. Four required tracheotomy at birth to one month of age. There have been no deaths. None of the four has had sufficient return of laryngeal function to permit extubation. Four of the seven have other central nervous system lesions; one is a mongol and three had central nervous system birth injuries; one of the seven had an associated subglottic stenosis and two were apparently normal except for the laryngeal paralysis.

Of the four infants with total left cord and partial right cord paralysis, two male, two female, two had congenital heart disease and two were normal except for the laryngeal lesions. None required a tracheotomy; all are living.

Unilateral Paralysis. In 21 of the 63 infants of this series, the paralysis was unilateral, with involvement of the right cord in eight and the left in thirteen. In four of the eight cases of right cord paralysis, this finding was an isolated condition causing a weak, breathy cry in an otherwise apparently normal infant. In three cases, other neurologic findings were present; pharyngeal paralysis in one, a right facial paralysis in one, and a series of anomalies in the other, including a cleft palate, a right facial paralysis, Erb's Palsy, and a congenital œsophageal atresia with a tracheo-œsophageal fistula. In the eighth case, a large cystic hygroma was present on the right. Only one infant had to be tracheotomized at one month of age, and was extubated sixteen months later.

Nine of the thirteen infants with left cord paralysis had congenital heart disease, or anomalies of the great vessels. A large auricle and an interventricular septal defect was present in five of the nine, and a

sixth infant also had a large heart but the anatomical diagnosis of the anomaly was not completely established. One infant had a patent ductus, another a Tetralogy of Fallot and the ninth, a vascular ring. The association of the left vocal cord paralysis with congenital heart disease is quite apparent and undoubtedly accounts for some of the post-surgical respiratory problems in these patients. One of the thirteen required a tracheotomy.

Of the remaining four infants with left cord paralysis, one had multiple congenital anomalies, including other cranial nerve involvement; one had been operated for a congenital œsophageal atresia with a tracheo-œsophageal fistula; one had an associated laryngomalacia; and in only one, was the left cord paralysis an isolated finding in an otherwise normal infant.

The indications for the laryngeal examinations in the newborn which revealed the laryngeal paralysis herein recorded were dyspnœa, an absent or weak cry, abnormalities of deglutition, or known cardiac, central nervous system or œsophageal anomalies. The use of an anæsthetic for the laryngoscopy is unnecessary; it adds to the hazard of the procedure and makes difficult an evaluation of cord movement. Vocal cord action and a comparison of the motility of one cord with the other is best determined when the laryngoscope blade is introduced into the vallecula and the tongue lifted forward. Only after laryngeal movement has been studied during this phase of the examination should the laryngoscope blade be inserted under the epiglottis and the configuration of the cords, the anterior commissure and the subglottic surface studied. It is often apparent at this time that the blade of the laryngoscope fixes the right vocal cord through pressure on the right aryepiglottic fold. Such pressure-fixation simulates the appearance of right vocal cord paralysis and may lead to a false diagnosis if not re-evaluated with the blade in the vallecula to permit freedom of cord movement. Inspection must include a sufficient observation time to insure visualization during inspiration as well as during phonation or cry. Often this is difficult because of cyanosis, apnœa, or the cardiac status of the infant, the examination having been requested because of a known severe congenital cardiac anomaly. The administration of oxygen throughout the direct laryngeal examination is often necessary and the duration of the procedure itself must be limited or discontinued if cyanosis develops. Preparations for a tracheotomy should be made before beginning the laryngoscopy if dyspnœa is severe; not because of the laryngoscopic examination itself, but because the pathology disclosed by the examination; a bilateral paralysis may require a tracheotomy if life is to be maintained.

From these studies it seems apparent that laryngeal nerve paralysis in infants is probably more common than previously recorded. In a review of 63 infants with laryngeal paralysis, partial or complete bilateral paralysis

was observed in 42. In 21 of these the laryngeal paralysis was only one phase of a major neurologic disorder. In bilateral paralysis, the cry is clear, although weak, but the dyspnœa was severe enough to have required a tracheotomy in twenty of the forty-two cases. Dysphagia with aspiration of food was a common phenomenon because of the frequency of an associated pharyngeal paralysis. Unilateral vocal cord paralysis was

FIG. 8. (*a*)
A.—Duplication of the right upper lobe bronchus.

observed in 21 of the 63 infants; the right cord in eight, the left cord in thirteen. In only two of the 21 infants with unilateral paralysis was a tracheotomy necessary. The prominent clinical feature of the 21 was the weak or absent cry. In four of the eight infants with right cord paralysis, this paralysis was an isolated finding in otherwise apparently normal infants. However, in nine of the thirteen infants with left cord paralysis a congenital cardiovascular lesion was considered to be the ætiologic factor of the paralysis; five of the nine had a large congenital heart with an interventricular septal defect. Thus in infants with bilateral vocal cord paralysis the lesion is most frequently a part of a generalized central

nervous system disorder and as such, the prognosis is relatively poor. Infants with the right cord paralysis may have no other neurologic lesion or the condition may be associated with other peripheral anomalies; the prognosis for ultimate voice function is relatively good. In the majority of infants with left vocal cord paralysis an associated cardiovascular anomaly is the ætiologic factor. While the prognosis regarding voice is

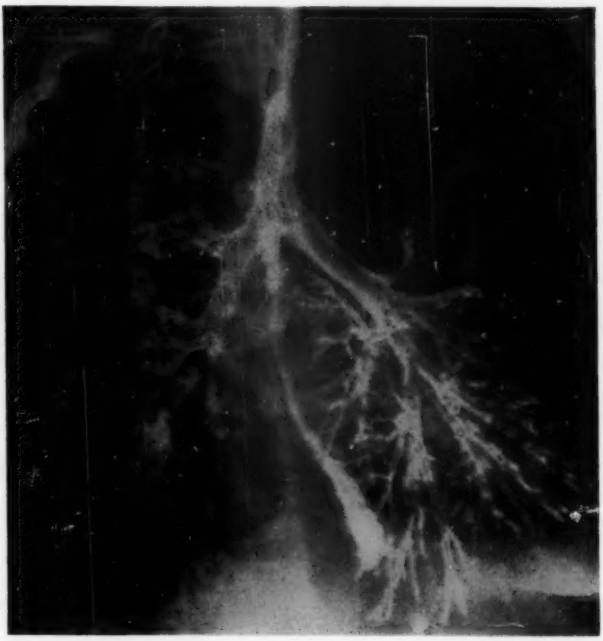


FIG. 8. (*b*)

B.—Accessory left upper lobe bronchus. Both the cyst and the anomalous right upper lobe and the emphysematous accessory left upper lobe were resected because of repeated pulmonary infections.

fair, the ultimate prognosis is dependent on the cardiac status and the presence of the cord paralysis may influence any contemplated surgical corrective procedure.

The Trachea and Bronchi

A didactic review of the congenital anomalies of the lower respiratory tract and the œsophagus will not be attempted in this discussion. However, of interest to the otolaryngologist are new endoscopic aspects of the

anomalies that have developed as thoracic surgery in infants has progressed. Many infants with anomalies heretofore invariably fatal survive the reparative surgery but require post-operative endoscopic management. In others, diagnosis and surgical therapy are directly

FIG. 9. (*a*)
The bronchogram of an infant with multiple congenital cardiac, genito-urinary and respiratory tract anomalies.

dependent on endoscopic findings which formerly might have been of only academic interest. A classification of anomalies, based on that of Jordan (1939), was suggested in a previous communication (Holinger *et al.*, 1956). Clinically, from the standpoint of the endoscopist, the problems are related to excessive tracheal or bronchial secretions, or

bronchial obstruction which has resulted in obstructive emphysema or atelectasis.

Apparent atelectasis of an entire lung in the newborn usually is found to be an *agenesis* rather than an actual atelectasis due to an obstructed bronchus. We have observed nine infants and one seven-year-old boy with this anomaly (Oyamada *et al.*, 1953). Eight of the ten were female; the lung was absent on the left in five and on the right in five. Six were referred by the pædiatrician or obstetrician after tracheal catheterization

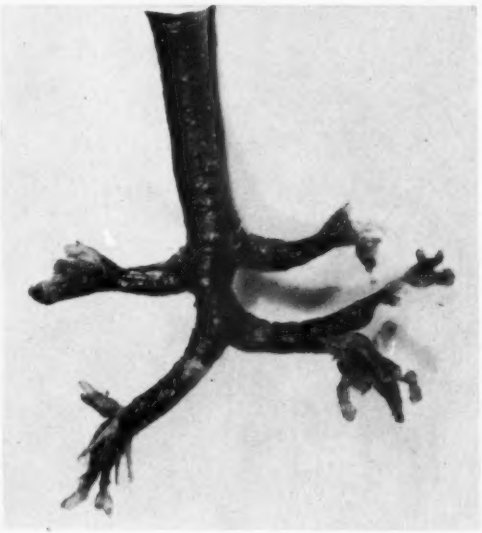


FIG. 9. (*b*)
The tracheobronchial tree of an infant with multiple congenital cardiac, genito-urinary and respiratory tract anomalies.

attempts had failed to remove the suspected bronchial mucus plug. The remaining four were seen at three months of age or older suspected of having an atelectasis due to food or other foreign bodies. Occasionally a small bronchial stump represents the original bronchial bud but even this may be entirely absent (Fig. 7). The condition is not incompatible with life although often one such severe anomaly is associated with others of equal severity in other systems. Duplications of parts of the bronchial tree, usually merely incidental findings during bronchoscopic examinations may be associated with conditions of real clinical significance in the newborn. The illustrations show a cyst in a duplication of the right upper lobe of one infant and an emphysema in an accessory left upper lobe of a child with a long history of repeated respiratory infection (Fig. 8). A third

bronchogram illustrates both a tracheal right and left upper lobe bronchus, the trachea continuing in the midline and then dividing to form a middle and lower lobe on each side (Fig. 9). The infant had a dextrocardia and numerous other anomalies ultimately incompatible with life. In another infant, referred for bronchoscopic aspiration of multiple lung abscesses of

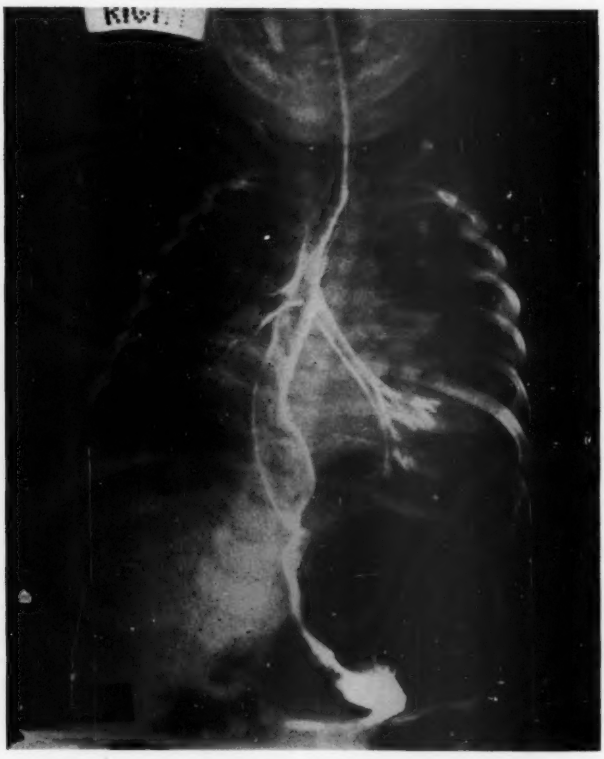


FIG. 10. (*a*)

A.—Bronchogram of an infant with multiple lung abscesses. Note the abnormally long left main bronchus.

the left upper lobe, it was found that these were in an accessory lobe originating from the œsophagus (Fig. 10). Tracheal or bronchogenic cysts, pinched off pockets of bronchial epithelium, likewise produce tracheal or bronchial obstruction requiring resection often in the first weeks of life (Fig. 11). Endoscopically they differ from the vascular anomalies in that they are much more firm and resistant to the advancing scope than are the soft compressible vessels.

Anomalies of Larynx, Trachea, Bronchi and Œsophagus

The phenomenon of *obstructive emphysema* in the newborn infant is one of particular challenge in infant bronchoscopy. Congenital lung cysts (Evans, 1949), a bronchial web, vascular anomalies compressing the bronchi (Holinger *et al.*, 1958) or an idiopathic lobar emphysema (Fischer *et al.*, 1952) may give identical physical and X-ray findings in the new-

FIG. 10. (*b*)

B.—The œsophagram demonstrating the œsophageal origin of the accessory abscessed lobe.

born. Bronchoscopic inspection is essential prior to surgical exploration. The web illustrated in Fig 12*a* is similar to that seen in the left bronchus of a cyanotic one-day-old infant whose X-rays are seen in Fig. 13. Dilatation of the web resulted in prompt relief of dyspnœa and a return of the heart and mediastinum to their normal position. The recognition of the absence of intrabronchial obstruction and extrabronchial compression in the extremely dyspnœic one-week-old infant whose X-rays appear in

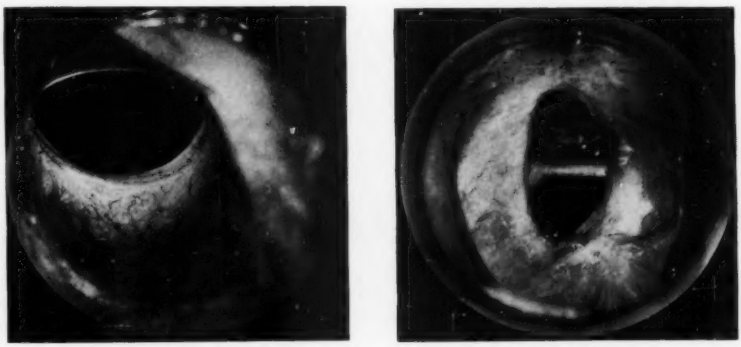
Fig. 14 permitted an immediate, life saving left upper lobectomy for an emphysema almost identical to that of the previous infant. Although the clinical findings and X-rays were almost identical in the two infants, the demonstration of the bronchial web in the first and the absence of it in the second made possible a correct pre-operative evaluation of the indications for or against an exploratory thoracotomy.

FIG. 11.
Tracheal cyst in the mediastinum causing severe respiratory obstruction.

These cases have been reported in detail previously (Holinger *et al.*, 1956) and are mentioned here to stress the significance of atelectasis and emphysema in the newborn. Professor Eelco Huizinga (1955) in his Semon Lecture for 1954 reviewed these phenomena as they occur in adults and correlated the pathology with that of other parts of the respiratory tract possibly of more immediate interest to most otorhinolaryngologists.

However, the field broadens in infants as we review another area of interest to the laryngologist, pædiatrician and the thoracic surgeon.

Obstruction of the trachea and œsophagus by anomalies of the great vessels. During the past fifteen years this has become one of the most important clinical problems associated with congenital anomalies of the tracheobronchial tree and œsophagus (Holinger *et al.*, 1948). Congenital malformation of the vessels originating from the branchial arches may produce severe respiratory and œsophageal obstruction (Fig. 15). The anomalous development or failure of resolution of portions of the arches results in partial or complete encircling constriction of the trachea and



(*a*) FIG. 12. (*b*)
Congenital tracheal webs.

œsophagus, or occasionally, of either one alone. Most commonly these anomalies involve the aorta and its branches, but in rare instances the pulmonary arteries separately or in conjunction with other structures are responsible for the respiratory or œsophageal obstruction (Potts *et al.*, 1954).

During the past twelve years, 41 infants or children have been seen at The Children's Memorial Hospital with vascular anomalies causing enough compression of the trachea, a bronchus or the œsophagus to require surgical treatment (Riker, 1954). It is significant and pertinent to this discussion, that a high percentage of the infants with these anomalies were referred by pædiatricians to the Division of Laryngology and Broncho-œsophagology because of their respiration or deglutition symptoms. Respiratory symptoms varied from simple inspiratory stridor to severe inspiratory and expiratory obstruction with acute cyanosis (Fig. 16*a*). All symptoms were increased in severity during feedings and respiratory obstruction generally precluded any activity. The wheezing, retractions of the sternum, dyspnœa and cyanosis were not always

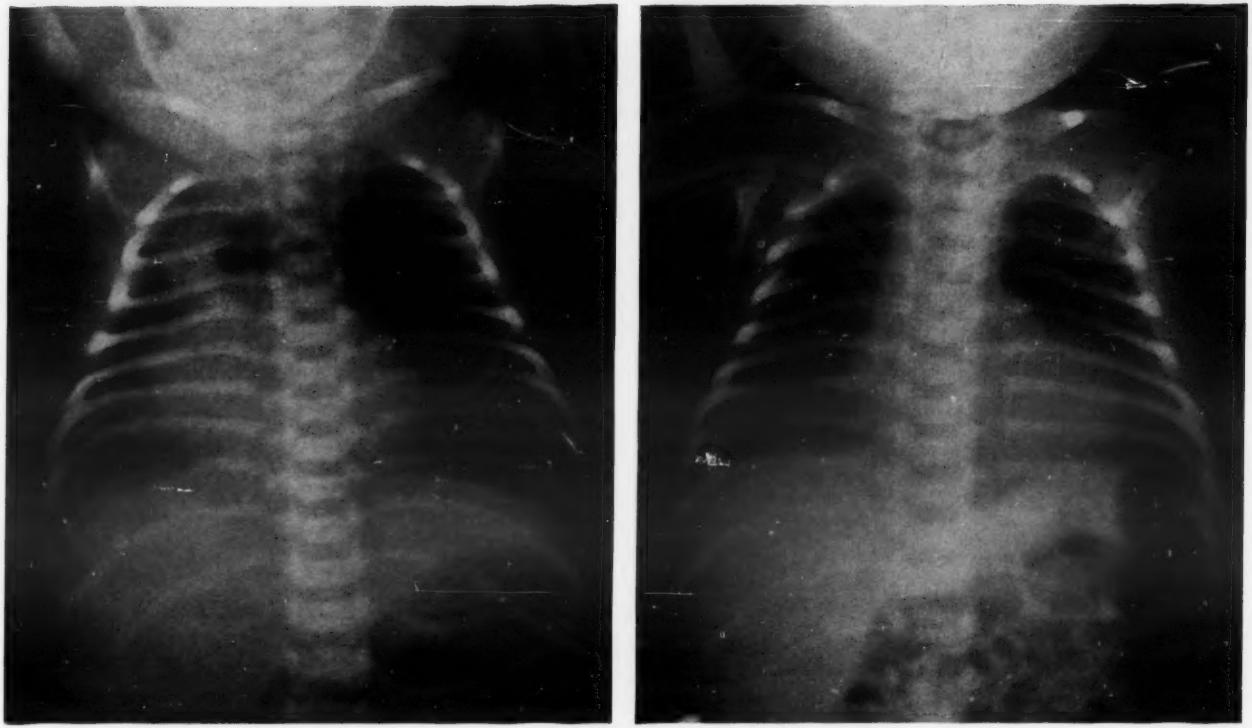


FIG. 13.

A.—Obstructive emphysema of left lung of one-day-old infant due to congenital web of left bronchus similar to that seen in Fig 12A.

B.—Heart and mediastinum in normal position twenty-four hours later after bronchoscopic dilatation of the pinpoint opening through the web.

the same in spite of the fact that the anatomical defect was constant. A persistent, brassy cough, sometimes referred to as a "seal bark" was present in most of the infants and repeated attacks of "croup" were noted in the histories of the infants and children over a few months of age. The œsophageal symptoms consisted of varying degrees of dysphagia, re-

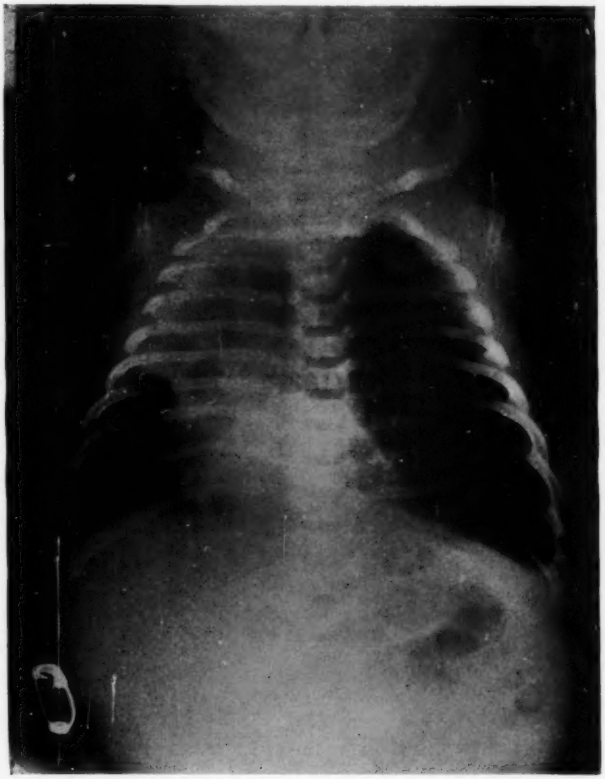


FIG. 14. (*a*)
Idiopathic emphysema of the left upper lobe. An immediate lobectomy was performed following bronchoscopy which had demonstrated a normal left bronchus

gurgitation and "feeding problems". While in some, these symptoms were present from birth, in many they were first noted at four to five months of age when semi-solid and solid foods were added to the diet.

Of greatest diagnostic importance is the œsophageal fluoroscopy which should be done only when the infant is hungry since then the infant eagerly swallows sufficient barium to permit adequate inspection (Figs. 16*a* and *b*).

A posterior indentation of the œsophageal barium column is the significant X-ray finding, but occasionally this is absent and the endoscopic findings are of greatest importance. Bronchoscopy assists in locating the obstruc-

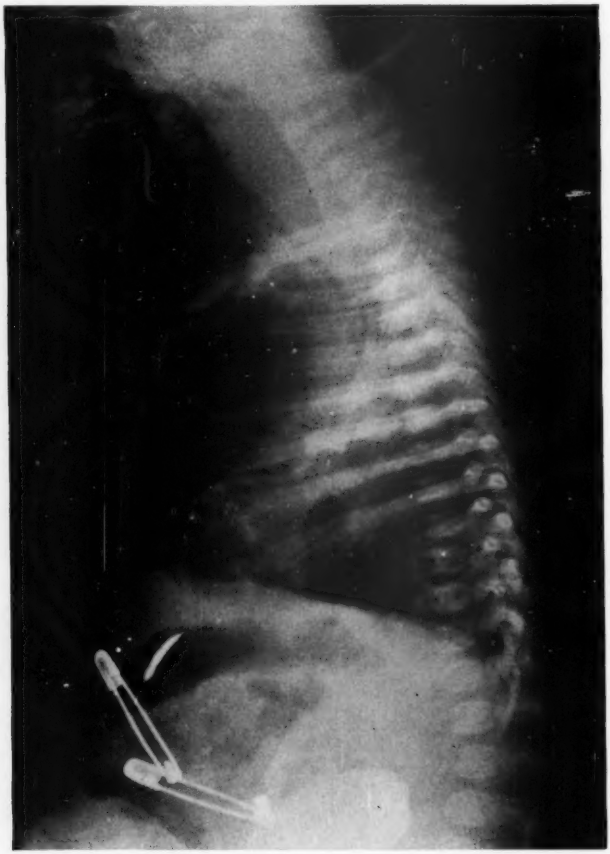


FIG. 14. (*b*)

Idiopathic emphysema of the left upper lobe. An immediate lobectomy was performed following bronchoscopy which had demonstrated a normal left bronchus.

tion as well as establishing the degree of change in the tracheal or bronchial wall. Typical findings are an antero-posterior tracheal flattening at the level of the aortic arch apparently due to extrinsic compression. The bronchoscope in most cases, can be advanced beyond this obstruction with ease, resulting in improved respirations. Many times a large accumula-

tion of secretion is found below the obstruction. Œsophagoscopy is also significant and occasionally the actual vessel involved can be identified by palpating the wrist and carotid pulses as the œsophagoscope compresses the vascular indentation (Samson and Dugan, 1951). The presence of extra-œsophageal pulsation is not easily established since all mediastinal structures in the infant undergo considerable motion normally during any endoscopic examination. Bronchograms were not used

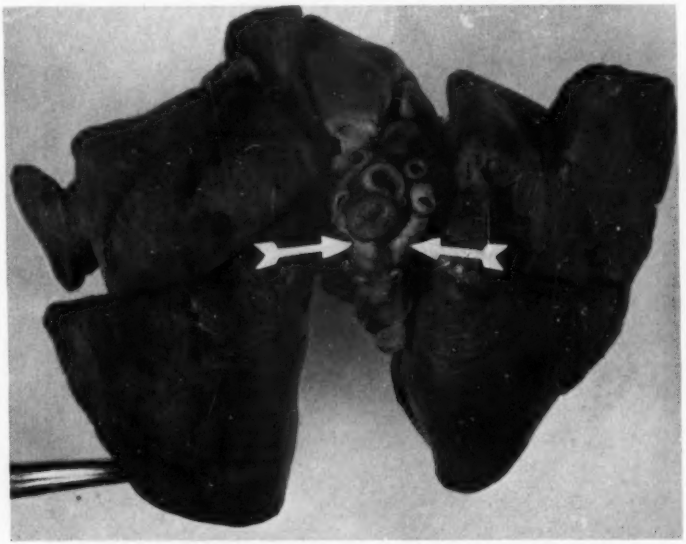


FIG. 15.
Double aortic arch responsible for asphyxial death.

routinely in the diagnostic studies but were done in cases in which there was some question as to the degree of involvement or its exact location. Post-operative bronchography revealed interesting findings of persistent anatomical narrowing of the affected viscus in spite of relief of the pressure of the anomalous vessel.

Consideration of other causes of intrinsic and extrinsic tracheal or bronchial obstruction is essential. Mediastinal tracheal or bronchial cysts or tumours and intratracheal webs or cartilaginous deformities may give symptoms simulating those of vascular rings. Laryngoscopy, bronchoscopy and œsophagoscopy assist in eliminating other laryngeal or tracheobronchial lesions from consideration. The possibility exists of increasing the obstruction by these diagnostic procedures only if extreme com-

pression is present. However, under these circumstances, the diagnosis by X-ray is usually obvious and bronchoscopy can be dispensed with.

Of the 41 infants and children operated with vascular compression of

FIG. 16. (*a*)

A.—Lateral chest X-ray showing tracheal compression due to vascular ring with a large anterior aortic arch.

the trachea, bronchi or œsophagus, 23 were males, 18 females. The age at the time of operation ranged from three weeks to ten years. Twenty-eight were under one year of age at the time of operation and thirteen were operated between the ages of one and ten years.

A simple, practical classification divides the patients into two groups:

Anomalies of Larynx, Trachea, Bronchi and Œsophagus

I. Those with complete vascular rings in whom both arches persisted.

II. Those with persistence of only one arch but with incomplete rings made up of constricting surrounding vessels or remnants of vessels (Fig. 17).

FIG. 16. (*b*)
B.—Œsophagram of infant with large posterior arch.

I. *Complete Rings.* This anomaly was found in twenty-two of the patients operated. It consists of a persistence of both aortic arches and is associated with the severest symptoms. The ligamentum arteriosum has always been on the left. There were five immediate post-operative deaths in the twenty patients in this group, usually caused by respiratory complications. The anomaly is found in three principal variations.

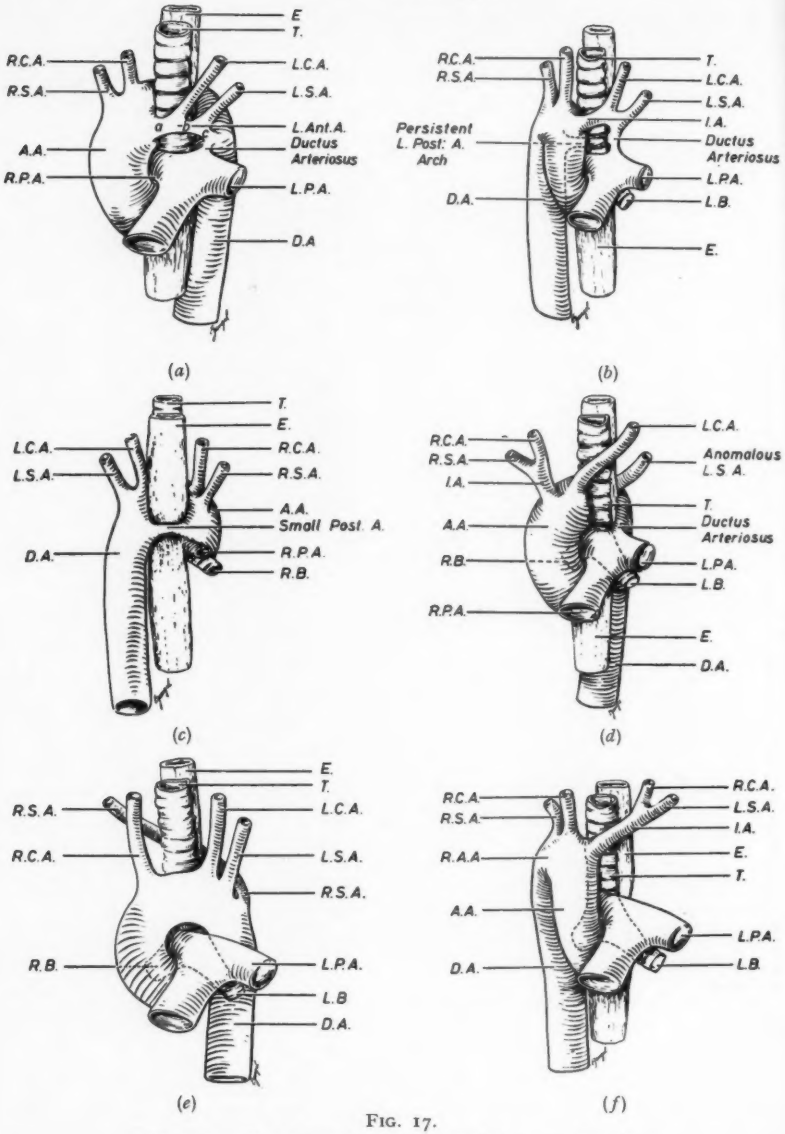


FIG. 17.

Diagrammatic representation of constricting vascular rings.

A.—Small left (anterior) arch; *a*, *b*, and *c* possible points for sectioning.

B.—Similar to A, left (anterior) arch small but aorta descends on right.

C.—Large left arch (anterior segment), small right (posterior) segment; aorta descends on left.

(*a*) That in which the left arch or anterior segment is small and the aorta descends on the left (Fig. 17*a*). Nine patients with this anomaly were operated and there were no deaths.

(*b*) Similar to (*a*), with the left arch or anterior segment small, but the aorta descending on the right (Fig. 17*b*). There were seven infants in this category that were operated with two deaths, both respiratory. It is apparent that in these patients the descent of the aorta on the right tightens the ring and increases the severity of all symptoms, particularly those of respiratory obstruction.

(*c*) The anomaly of a large left arch or anterior segment. While it is possible that in these patients the aorta could descend on either side, the descending aorta was found on the left side in each of the six cases seen with this type of anomaly (Fig. 17*c*). There were three post-operative deaths in the six patients in this group.

II. *Incomplete Rings*. Vascular compression of the air or food passages due to incomplete vascular rings may be due to many combinations of vessels and rudimentary structures. In this group, only one aortic arch persists. There were sixteen patients in this group operated with three deaths. The incomplete rings may also be divided into two principal categories:

(*a*) A right aortic arch with a left-sided ligamentum arteriosum (a remnant of the sixth arch) (Fig. 17*d*). In this category the aorta may descend on the right or the left to further subdivide this group. There were eleven patients operated whose defects were of this type with two deaths. Both were found to have interventricular septal defects.

(*b*) A ring formed by an anomalous origin of the right subclavian artery originating from the left descending aorta below the origin of the left subclavian artery (Fig. 17*e*). There were five patients operated with this type of lesion with one death. This infant also had Eisenmenger's Disease and a cleft palate.

D.—Right aortic arch with left-sided ligamentum arteriosum.
E.—Right subclavian artery originating from left descending aorta (Dysphagia Lusoria).
F.—Uncomplicated right aortic arch and right descending aorta. This may produce severe compression of the right main bronchus.

T—Trachea
E—Œsophagus
B—Bronchus
SA—Subclavian artery
CA—Carotid artery
PA—Pulmonary artery
AA—Ascending aorta
DA—Descending aorta
IA—Innominate artery
Ant. A—Anterior arch
Post A—Posterior Arch
R—Right
L—Left

Three cases constitute a miscellaneous group. In one the innominate vein apparently produced pressure on the trachea and symptoms were relieved by lifting it and attaching it to the sternum. Further relief was obtained after removing the thymus of this patient. Obstruction was

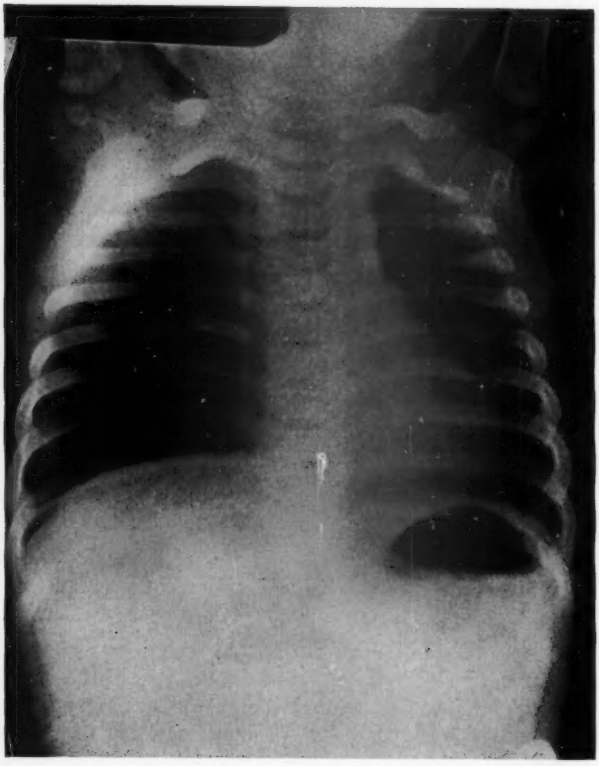


FIG. 18. (*a*)

A.—Obstructive emphysema of the right lung produced by an anomalous left pulmonary artery which coursed around the lower trachea and right bronchus before going to the left lung.

considered due to the combination of an enlarged thymus and the anomalous innominate vein. In the second patient, a large, somewhat anomalously placed azygos vein constricted the œsophagus to cause dysphagia. In the third patient (Fig. 18) severe obstruction of the right bronchus was found to be caused by an anomalous left pulmonary artery which coursed around the right bronchus before going to the left lung

(Potts *et al.*, 1954). There was no operative mortality in these three patients.

That a right aortic arch and right descending aorta (Fig. 17*f*) may cause severe obstruction of the right main bronchus has been discussed

FIG. 18. (*b*)

B.—Bronchogram showing polyethylene tube supporting trachea and right bronchus following resection and re-anastomosis of left pulmonary artery.

elsewhere (Holinger *et al.*, 1948). This anomaly caused a fatal obstructive emphysema in an infant prior to the time corrective surgery was considered although the bronchial compression and its probable vascular cause were recognized. In addition to the compression, an abnormally long right main bronchus was found, with the origin of the right upper lobe considerably below the level of the carina.

The surgical correction of vascular anomalies compressing the air or food passage should be undertaken as soon after the diagnosis has been established as possible. This is not only to prevent possible sudden asphyxial death, but to take full, effective advantage of the early growth

FIG. 19. (*a*)

A.—Congenital œsophageal atresia without fistula. Note the absence of air below the diaphragm.

potential of the constricted trachea. Kirklin and Claggett (1950) suggest that prolonged pressure on the trachea can lead to softening of the tracheal rings. It is our experience that the rings fail to develop originally, due to the pressure of the anomalous vessel during growth in the embryo

and in fœtal life. This was demonstrated in infants in whom these anomalies were recognized prior to the time that surgical intervention was considered. In either case the earlier the pressure is removed, the more opportunity there is for normal growth to be fully effective. Final improvement in symptoms often was delayed until the tracheal cartilages could

FIG. 19. (*b*)

B.—Foreign bodies, buttons, in a colon transplant serving as a substitute œsophagus in a child who had had an atresia of both segments of the œsophagus.

grow to their normal rigidity and configuration. There have been nine deaths among the 41 patients operated. Most of these had other severe complicating or disassociated congenital anomalies. Some of these anomalies were incompatible with life. Most of the deaths were in young infants with severe low tracheal obstruction. Tracheotomies became necessary because of the œdema and constriction at the level of the tracheal bifurcation. Under these circumstances, death occurred in some of the earlier cases because of erosion of the tracheal wall or the carina

itself by the tracheotomy tube, or because of obstructing granulations that formed around the end of the tube. Later this technique was altered in such cases to the use of a short tracheotomy tube and the advancement of a thin-wall polyethylene tube into the right bronchus below the

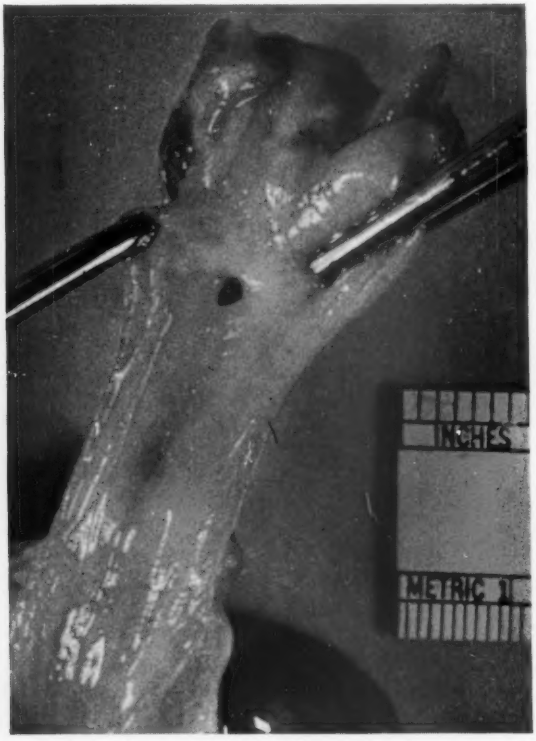


FIG. 20.
Tracheo-œsophageal fistula immediately below the cricoid cartilage. Extreme debility had prevented surgical closure.

obstruction. Holes were placed in the tube to permit ventilation of the left lung. This technique was effective and practical (Fig. 18*b*).

The Œsophagus

Congenital anomalies of the œsophagus have always been of interest to the laryngologist and in the past endoscopic management of the problems of the stenoses was almost exclusively a responsibility of the

departments of Otolaryngology. With the technical advances in thoracic surgery, this field has expanded as life-saving operations have added a large group of patients with new endoscopic needs. Pre-operative diagnosis and preparation, and the recognition and management of post-operative complications require the closest co-operation between thoracic surgeon

FIG. 21.
Cardiospasm in a newborn infant.

and laryngologist. Sensory, as well as motor paralyses of the larynx must be differentiated from tracheo-œsophageal fistulae; strictures of the trachea as well as the œsophagus may follow repair of an œsophageal atresia; and the management of the foreign body in a colon serving as an œsophagus, are surely the responsibility of the laryngologist interested in endoscopy as part of the broad field Sir Felix Semon envisioned.

No challenge is greater in numbers or variety than those of congenital

œsophageal atresia and fistulae; either may be seen alone but more commonly, of course, they are combined. During the past fourteen years, from 1946 to 1960, 195 infants with these associated congenital anomalies were operated at The Children's Memorial Hospital in Chicago by Dr. Willis J. Potts and his surgical staff (1960). These cases and others referred for endoscopic management after similar surgery at other institutions constitute a series not alone of statistical significance, but one which includes a fair percentage of the possible complications of interest to the laryngologist (Holinger *et al.*, 1951).

Of the 195 infants operated, atresia of the upper segment and fistula between the lower segment and the trachea or a bronchus was the anomaly seen in 169. Atresia of both upper and lower segments without fistula was seen in 17 infants (Fig. 19). Fistula without atresia was seen in six; and atresia of the œsophagus with communication into the trachea of both upper and lower segments was seen in three. The overall mortality in the 195 cases operated is 44 per cent.; this figure was higher during the first years but has fallen to approximately 30 per cent. for the past few years. The causes of death at the present time are prematurity, severe anomalies of other systems, or pneumonic complications. Of the 169 infants operated for an œsophageal atresia and tracheo-œsophageal fistula, more than half are living. It should be noted that approximately 5 per cent. of the 169 infants had right aortic arches. This always indicates the presence of Tetralogy of Fallot or a vascular ring. Under these circumstances the anastomosis of the two œsophageal segments is made to the right of the aorta, freeing the œsophagus and leaving ample room for the trachea inside the ring. Pre- and post-operative bronchoscopic aspirations frequently were requested in the early cases of the series but this need has decreased as technics of anæsthesia have improved and post-operative catheter aspiration has come to be used routinely and effectively. Most essential is that no oral fluids are given the first five days and even then are withheld until after fluoroscopy shows the condition of the anastomosis site. Particular care in feeding the infant slowly reduces the need of post-operative bronchoscopic aspiration. Dilatations with mercury-filled bougies, œsophagoscopic bougies or the œsophagoscope itself are begun ten days post-operatively if the lumen through the anastomosis is small. Early dilatations are important and the response more rapid than later dilatation when the stenosis is more fibrous. Furthermore, it is much easier to maintain the lumen than to re-establish it. Gastrostomy is not done routinely but only if a leak is demonstrated, if the infant is premature and too weak to suck, or if the infant was admitted for operation at more than a week of age.

Approximately half the infants that survive require dilatation procedures of one type or another. There has been a gradual reduction in the need for these procedures in our own series as technics of suture of the

anastomosis site improve. Those operated elsewhere who are referred for dilatation of the anastomosis are generally from four to five months of age, when semi-solids and solids are added to the diet and the stenosis becomes apparent. These are more difficult to manage and may require a gastrostomy and either retrograde or prograde dilatation using a thread guide. As these infants grow older, foreign body problems are common; not only food, but every type of object an infant or child might swallow (Holinger *et al.*, 1949).

Infants with complete atresia of both upper and lower œsophageal segments require an œsophagotomy and gastrostomy as the initial procedure since the segments are too short for primary anastomosis. In eight, a substitute œsophagus has been constructed using the colon in six, the jejunum in one and the stomach in one; there was one death during revision of the anastomosis site of one of the colon transplants. These children are observed with periodic post-operative barium studies and dilatations, usually over a thread, are instituted if strictures begin to form. Delay results in a stenosis that sometimes requires re-operation and re-anastomosis. The foreign bodies that must be removed from the jejunum or colon of these children by means of the œsophagoscope present additional problems of more than passing interest (Fig. 19*b*).

Tracheal stenosis was a serious complication in some of the early cases of atresia of the upper œsophageal segment and fistula between the trachea and lower segment. It is still a complication occasionally seen in infants initially operated elsewhere and later referred for endoscopy, indicating that its pathogenesis is not realized. The stenosis occurs in the region of the tracheal bifurcation or the entrance to the left bronchus at the site from which the lower segment has been removed. It is due to too tight a closure of the trachea, particularly if the fistula was a large one. Granulations accentuate the obstruction, necessitating dilatations with a 3 mm. or a Negus suckling bronchoscope to maintain the lumen. Those that require a tracheotomy for low tracheal obstruction generally have a poor prognosis because if the tube has to advance through the obstruction to provide an airway it touches the carina causing ulceration and more granulation tissue obstruction. This complication must be avoided at the time of the original operation; but if it occurs, early tracheal dilatation should precede any œsophageal dilatation procedures in order to avoid asphyxia as the œsophageal dilators compress the trachea.

Congenital Tracheo-oesophageal Fistula. In recent months we have become more aware than we were formerly of the frequency of this anomaly and its significance in regard to aspiration pneumonia. The image intensifier and cine-fluorography provide a much better means of diagnosis of œsophageal conditions in infants than is possible by more routine technics. Differentiation between a tiny, high fistula (Fig. 20) and aspiration due to an inco-ordination of the muscles of deglutition is a diagnostic

problem requiring the closest endoscopic and radiologic co-operation (White and Kraus, 1957). However, fistulae between the mid-thoracic œsophagus and the trachea are rarely endoscopic problems as they were in the past. Once demonstrated they are, of course, closed surgically.

Congenital Anomalies and Foreign Bodies in the Oesophagus. It is particularly important in this discussion to stress the major ætiologic rôle that congenital anomalies of the œsophagus plays in the lodgement of foreign bodies in the œsophagus (Holinger *et al.*, 1951). There are four types of congenital œsophageal lesions which should be considered; congenitally short œsophagus, congenital œsophageal stenosis, cardiospasm in infants (Negus, 1936) and strictures of the œsophagus which may follow the repair of a congenital œsophageal atresia (Fig. 21). The fourth type is self-evident and has been mentioned. By the time the infant is old enough to swallow solid food or foreign bodies, the physician and family are well aware of the œsophageal lesion and its problems, and understand the signs and symptoms of obstruction. They return the child to the hospital for removal of the obstructing food, and dilatations of the stricture are carried out to permit the child to resume normal feeding. Rarely is there any question of diagnosis. However, infants or children with a congenitally short œsophagus or a congenital œsophageal stenosis usually present no symptoms during the first five months of life. As infants they are on a completely liquid diet and fluids pass through the stenotic area without appreciable hesitation. When semi-solid foods are added to the diet, feeding problems manifest themselves by slow eating and a reluctance to take anything but liquids. Often the family becomes accustomed to the length of time it takes to feed such an infant but the child is sooner or later labelled a "feeding problem". When solids are added to the diet, frequent episodes of temporary obstruction occur with drooling, vomiting of undigested food and loss of weight. The child soon realizes he must refuse solid food entirely, thus further leading to the supposition that he is a "feeding problem". Bizarre eating habits develop, with the result that as the child grows older the time used to consume a meal may extend to an hour or more. Finally episodes of complete obstruction occur which require œsophagoscopic removal of a foreign body—a piece of meat, an orange seed, a toy or a coin—and then diagnosis is established. Thus, it is important that if a foreign body lodges in any part of the œsophagus other than the cervical œsophagus, its removal should be followed by fluoroscopic studies.

In closing I wish to thank the members of the Sir Felix Semon Lecture Committee for extending to me the privilege and honour of participating in this lecture series. Time and space do not permit consideration of the clinical significance of anomalies of other systems that secondarily affect

the air and food passages. The broncho-pulmonary suppuration associated with fibrocystic disease of the pancreas, the respiratory obstruction caused by tracheopathia osteoplastica and the œsophageal varices of infants with splenic or portal system anomalies are examples of but a few. I concur in the thoughts of Professor Huizinga who felt that the medical background of Sir Felix Semon justifies considering the subject matter presented here as being within Semon's concept of the field of Otolaryngology. While some otolaryngologists devote their attention to otology and others to rhinology, plastic surgery, allergy or to laryngology alone, still the closest ties between pædiatrics, general medicine and surgery—and otolaryngology are realized in the fields of broncho-œsophagology. The significance of this relationship in infants is growing; I hope that this field will continue to hold the interest of Otolaryngologists who developed it.

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700 N. Michigan Avenue,
Chicago, Illinois, U.S.A.

OTOMYCOSIS: A NEGLECTED DISEASE

By A. E. W. GREGSON and C. J. LA TOUCHE (Leeds)*

A SURVEY of British literature leaves the impression that otomycosis has never held a prominent place among ear diseases in this country. Dismissing the condition in a few lines, most authors suggest that it occurs infrequently and that treatment is a simple matter of applying some well-known preparation.

It is, however, apparent in most clinics that the discharging ear still presents unsolved problems. There is that small nucleus of patients who come and come again; whose treatment theme has infinite variations with but one tangible result—a steady increase in the thickness of their out-patient notes.

With these thoughts in mind the present writers decided, some four years ago, to investigate patients of this type for the presence of an active fungus infection. There were several good reasons for this. Local antibiotic therapy, while not in universal use, had attained significant proportions in the treatment of both external and middle-ear infections, and it has been recognized that antibiotic therapy elsewhere could lead to secondary infection by fungi. The very sparsity of British papers on the subject suggested that there might be much to learn about the rôle of the fungi in ear infections. Perhaps the most significant feature of the present series was the possibility of controlling clinical diagnosis by the investigations of a trained mycologist, and there were few examples of this having been done before.

From the start it was agreed that a diagnosis of otomycosis would be made only in those cases where a fungus was seen to be growing actively in a sample of debris taken from the external auditory meatus. Since fungus spores abound in the atmosphere, and might well enter the meatus only to lie dormant there, their detection by the culture of a swab taken from the meatus could not be regarded as evidence that they were playing an active part in an infective process therein. Where it appeared that the fungus was, in fact, playing an active part in infection it was planned to give treatment aimed specifically at eliminating it. Resolution of the clinical condition might then be accepted as reasonable proof that the fungus had caused it, or was a contributing factor in its maintenance.

It was, of course, understood that this proof did not necessarily satisfy Koch's postulates. No attempt to transfer the infection to animal

* From the General Infirmary at Leeds.

hosts was to be made. This had, however, been done already by other investigators. In any case, Whalen (1938) has commented on the unreasonable restriction imposed by rigid observation of the fourth postulate.

Results of the Investigation

During the years 1956-9 180 patients were examined, of whom 83 were found to have a fungus infection (Table I). Sixty-two of these patients had infections of the external auditory meatus and 21 had infections in radical mastoidectomy or fenestration cavities.

TABLE I.

NUMBERS OF PATIENTS EXAMINED FOR OTOMYCOSIS.

Year	Total examined	Total positive
1956	7	5
1957	15	8
1958	20	11
1959	134	59

TABLE II.

NUMBERS OF DIFFERENT FUNGI ISOLATED.

1956		1957	
A. flavus	2	*A. fumigatus*	1
A. niger	2	*A. niger*	3
Candida Sp.	1	*C. albicans*	2
Yeasts	1	*C. krusei*	1
		C. parakrusei	1
1958		1959	
A. fumigatus	1	*A. fumigatus*	13
A. niger	3	*A. niger*	12
A. nidulans	1	*A. nidulans*	1
Scopulariopsis Sp.	1	*A. terreus*	3
C. albicans	4	*A. herbariorum*	1
Candida Sp.	1	*C. albicans*	12
		C. parakrusei	1
		C. guillermandi	1
		A. niger and *C. albicans* ..	1
		A. candidus Sp.	1
		Yeast Sp.	10
		S. brevicaulis	1
		Actinomycetes	2

The fungi found were predominantly species of Aspergillus or Candida (Table II). Although Aspergillus has long been recognized as a cause of otomycosis (Capps, 1938, Reeh, 1942, Johnston, 1944) there has been little reference to Candida previously. Even so, its occurrence in almost half of the cases should occasion little surprise for it is a well-known pathogen in other sites. It is, however, noteworthy that organisms such as Penicillium (Whalen, 1938) and Mucor (Johnston, 1944) do not appear in the series.

Clinical Picture

This was not known. Descriptions in books of reference were brief and uninformative. With observation, however, patterns have become apparent, but even now they cannot be offered as anything more than impressions, and clinical diagnosis is still uncertain and inaccurate.

One symptom has occurred with some constancy. This is a sensation of irritation in the meatus which may be almost intolerable. Where the fungus

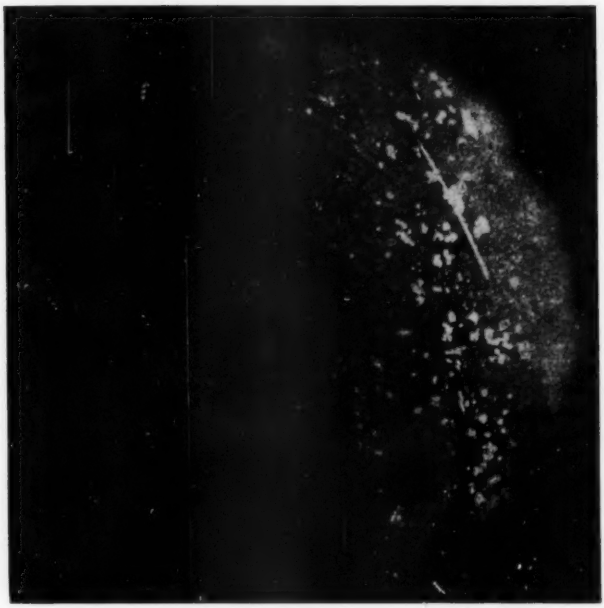


FIG. 1.
Clinical picture of infection by *A. flavus* as seen with the electric auriscope.

is causing otitis externa pain is unusual unless there is an associated pyogenic infection. Capps(1959) has, however, found that pain can be a symptom when the fungus causes infection in a radical mastoidectomy cavity and this observation has been confirmed. Discharge is scanty and is usually quite odourless. Unless some other factor causes a predisposition to it, deafness is not usual.

The meatus is rarely swollen unless self-inflicted trauma has led to a secondary pyogenic invasion. In the acute stage it is filled with a mass of grey caseous debris, quite unlike the "wet blotting paper" of classic descriptions. Removal of this debris shows a meatal lining of a distinctive

magenta colour, covered with small glistening white masses. The tympanic membrane, although discoloured, is of normal contour.

Less acute cases may present two patterns, depending on the type of infection. Where Aspergillus is present (Fig. 1) the meatus is notably dry and the magenta colour is less evident. Clusters of conidiophores may be seen, resembling miniature pin-heads. They may be white, yellow, brown, black, or green, depending on the age and species of Aspergillus. Simpson

FIG. 2.
Clinical picture of infection by *C. albicans* as seen with the electric auriscope.

(1959) has described this picture as an asymptomatic condition among native troops in India, but it has occurred only once in a European in this series.

In Candida infections the picture more nearly resembles the acute case after cleansing (Fig. 2). The meatus is generally moist and small white aggregations are scattered evenly throughout. In the plotograph they may be seen as wavy white lines radiating from the centre of the drum.

These features are not easily seen with the naked eye, but are quite apparent when the electric magnifying auriscope is used. Previous treatment may, however, modify the clinical picture out of all recognition, particularly if oily preparations have been used. The writers have, there-

fore been prepared to suspect a mycotic infection in any cases where there is a history of irritation or of previous antibiotic therapy, or where standard treatment, such as wicks of ichthyol in glycerine, has failed to produce a cure. In the interests of accurate diagnosis it is, of course, essential to have the confirmation of a direct mycological examination in these more doubtful cases.

Ætiology

Previous failure to clarify the clinical picture may have much to do with the reason for the surprisingly high frequency of this type of infection today, but the ætiology of the condition remains obscure. It is doubtful if the fungi are primary invaders of the external auditory meatus, although Brown (1937) held that they were, and occasionally there is a striking illustration.

CASE I.

A. B., male, aged 35, had no previous history of ear trouble. He was an enthusiastic yachtsman, and while sailing on a fresh-water lake one week-end his boat capsized, most of its equipment being dropped in about eight feet of water. He spent some time diving to retrieve this equipment, but, unable to recover it all, went back to the boat-house and searched the sail-loft for a grapnel. During this search he moved much unused equipment with a considerable disturbance of dust. Some three days later he noticed slight irritation in the ears, and as this increased over the next few days he sought advice. On examination he had an obvious infection of the ear canal with Aspergillus which was identified subsequently as *A. flavus*.

In other cases it has been possible to identify a primary source of infection elsewhere on the patient.

CASE II.

Mrs. C. D. was some four months pregnant when she presented with Candida infection in both external auditory meati; this responded satisfactorily to treatment but soon relapsed. Investigation by her obstetrician showed Candida infection of the vagina which proved resistant to treatment. The ear infection continued to relapse, but cleared up within two weeks of her delivery when the vaginal infection settled also.

The rôle of bacteria in paving the way for fungus infection remains open to question. Not all the cases in this series were subjected to bacterial investigation when the diagnosis of otomycosis was first made. Where this was done, however, a wide variety of bacterial flora was found. Of 27 cases so examined, 4 gave sterile cultures, 16 grew organisms of the pyocyaneus, proteus, or coliform types, 5 showed *Staphylococcus aureus* and 2 produced "yeasts".

The exudate occurring in bacterial ear infections seems likely to

provide a degree of humidity favourable to fungal proliferation and it is clear that bacteria and fungi can co-exist in the same clinical condition. Antibiotic therapy then might suppress the bacteria primarily responsible, leaving the fungi free to grow. In the present series 30 cases had definite evidence of previous local antibiotic therapy, while in a further 31 cases evidence was less certain since therapy had been applied previously elsewhere and details were unavailable. Many of the Aspergillus species which were found are known to produce effective antibiotic agents in their

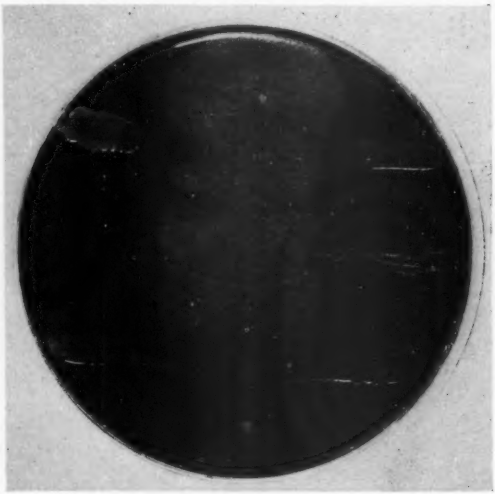


FIG. 3.

Culture plate showing the inhibiting effect of *A. fumigatus* on the growth of *Staphylococcus aureus* and *Streptococcus viridans*.

metabolism, although they may be too toxic for systemic use or have not yet been developed commercially. Fig. 3 shows the inhibiting effect of *A. fumigatus* on various known pathogenic bacteria. The failure to inhibit the growth of *Ps. aeruginosa* and *E. coli* is noteworthy.

A consideration of the metabolism of the common fungi found in the external ear can only occasion surprise that they are not invaders more frequently. Needing little more than moisture, warmth, a little protein or carbohydrate, and traces of mineral salts, their growth can be quite exuberant. Their proliferation in such unattractive sites as the space between the two lenses of a microscope objective is well known in tropical climates.

Nevertheless, examination of 32 clinically normal meati of volunteers from the hospital staff showed no evidence of actively growing fungus,

and the subsequent culture of swabs from these meati gave only 2 positive results. As these occurred in a technician in the mycology department and a nurse in the allied department of dermatology, they could have been transient contaminants.

This does suggest that the normal secretion of the meatus may have an inhibiting effect on fungal proliferation. Senturia (1957) quotes several authorities in saying that most of the saturated and unsaturated fatty acids have some inhibitory effect on the majority of fungi. He mentions Akobjanoff *et al.* (1954) who identified nine saturated and unsaturated fatty acids in ear wax.

Mycological Aspects

It is clear from the experience gained by the authors as a result of investigating the mycology of otomycosis that the fungi most prominently associated with this condition are mainly species of the genus *Aspergillus* and various yeast-like species belonging to the genus *Candida*. They, among so many thousands of fungi whose spores and hyphae contaminate the atmosphere, are able to grow in the external auditory meatus while, evidently, the vast majority of fungi are unable to do so. They alone are able to establish themselves in this site with success and frequency in the presence of large numbers of bacteria.

A necessary prelude to an inquiry into this problem is to consider first of all what is already known of the main physiological requirements for the growth of fungi.

Main Physiological Requirements for Growth

These are, in short: adequate moisture, adequate warmth and adequate nutrition.

While such requirements vary within given limits for different species, it may be stated that, by and large, a percentage relative humidity of 70 to 80, a temperature range of 15° C. to 30° C., and a minimum supply of certain chemical elements, viz., carbon, oxygen, hydrogen, nitrogen, phosphorus, sulphur, potassium, chlorine, magnesium, as well as traces of iron, manganese and of certain vitamins, represent the conditions necessary for growth in the case of the majority of fungi. Certain fungi, however, grow, not only well at temperatures lying between 30° C. and 40° C., but actually grow and reproduce more rapidly within this temperature range than at lower temperatures, and a few find their optima for these functions at even higher temperatures.

It is also evident that certain moulds are able to grow in an environment in which the oxygen tension is low and in which the carbon dioxide concentration is relatively high; although it is well known that carbon dioxide concentration is an important factor in controlling both growth

and reproduction in the fungi, especially when other conditions are minimal for growth. These effects of carbon dioxide concentration on fungus growth and reproduction have been studied by various authors but especially by Brown (1922).

Published data concerning carbon dioxide concentrations in the external auditory meatus appear to be lacking.

General and Selective Factors which may be Significant in enabling certain Moulds and Yeasts to become established in the External Auditory Meatus

It does not require experimental proof to show that the percentage relative humidity in the external auditory meatus is high, particularly when otitis externa is present, nor that the temperature in this site closely approximates "body temperature". Epithelial debris and serous exudate in various stages of chemical breakdown also provide a pabulum which is apparently suitable for moulds and yeasts to become established in the external auditory meatus when otitis externa is present. Such conditions should be, it might seem, suitable for many moulds and yeasts, even if not the best for particular species. Yet the fact remains that a particular group of moulds, namely certain species of Aspergillus, and a particular group of yeast-like fungi are predominantly favoured in these circumstances. To explain this, certain selective factors may be postulated. First of all, there is the factor of high temperature. All the species of Aspergillus whose active presence in the external auditory meatus was established microscopically and culturally in this series have been found subsequently to be able to grow and reproduce more rapidly at 37° C. than at 27° C., as judged by naked-eye observation within the first 24 hours following inoculation of culture slopes with a standard loopful of spore suspension. This is likewise the case with the species of Candida isolated in the series.

Another selective factor may be the ability of some of these species to produce antibiotics which enable them to establish themselves in competition with the bacteria which are almost invariably associated with them in otitis externa. A list of these antibiotics may be found in volume 1 of *Antibiotics* by Florey *et al.* (1949). From this list it may be seen that *A. fumigatus* produces several antibiotics, of which the most interesting in this connection may be fumigatin. This antibiotic is said to be active against certain strains of *Staphylococcus aureus* in glucose broth in a dilution of 1 in 50,000, and against *Escherichia coli* in a dilution of 1 in 20,000. Also certain strains of *A. niger* produce penicillin and likewise strains of *A. flavus* and *A. nidulans*. Citrinin, produced by *A. terreus*, is said to be particularly active against some Gram-positive organisms; for example, in a dilution of 1 in 24,000 it inhibits some strains of *Staphylococcus aureus*.

A third selective factor is certainly pathogenicity. There is no lack of records of the pathogenicity of *A. fumigatus* in other sites of the body and other species of Aspergillus such as *A. nidulans*, *A. flavus* and *A. niger* have been found from time to time associated with various lesions. The pathogenicity of *C. albicans* is also not in doubt; it has been recognized for over 100 years. In recent years a voluminous literature concerning the pathogenic activities of species of Candida and in particular *C. albicans*, in man and animals has accumulated. It is best known as the agent of thrush, but it is also a secondary invader in various forms of chronic pulmonary disease, a major cause of such common diseases of the cutaneous surfaces as paronychia, perionychia and onychia, intertrigo, rashes in the napkin area especially during the neonatal period, and of vaginitis and pruritus ani in adults.

Mycological Investigation of Patients Diagnosed Clinically as Cases of Otomycosis

The need for microscopic confirmation of a clinical diagnosis of otomycosis has been emphasized already. But the mycologist is only rarely to be found on a hospital staff, and the otologist may well have to perform these investigations himself if he is to achieve accurate diagnosis. This brief summary of the microscopic and cultural features of the Aspergillus and Candida groups is included as a convenient means of reference.

Superficial Examination. Debris suspected of being fungoid in nature was first located by use of the electric auriscope. By this means it was possible, sometimes, to see quite clearly the conidiophores of species of Aspergillus projecting into the canal from the surrounding walls, roof, and floor. Minute black heads at the tips of the sporulating stalks (conidiophores) indicated that *A. niger* was present. Likewise, pale blue or green plume-like growths (columns of conidia in chains) on stalks were indicative of *A. fumigatus*. Other species could not be identified so easily by this method, since their appearances were not exclusively characteristic. Thus, the light brown conidia of *A. terreus* may be confused with the immature conidial heads of *A. niger*, and those of *A. candidus* may be mistaken for the immature conidia of most of the commoner species of Aspergillus, since pigment develops in the conidia only as they become mature.

Signs suggesting the presence of Candida species, though by no means acceptable as diagnostic, were the presence of white or cream coloured viscous deposits on the epithelium of the external meatus.

Microscopical examination was necessary in all cases in order to confirm the preliminary diagnosis.

Bacteria. Bacteria of one kind or another were nearly always present in association with fungi, but the electric auriscope was of little use in

establishing their presence. When, however, a viscous discharge showed a greenish tinge, *Pseudomonas aeruginosa* was usually suspected, and this was often confirmed on microscopical examination after staining, as well as by culture. This, however, did not by any means exclude the presence

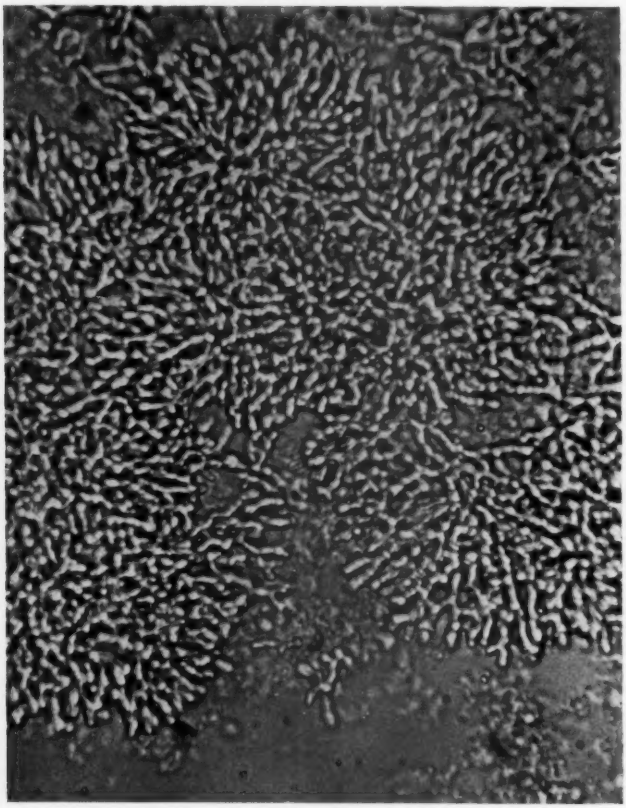


FIG. 4*a*.
Direct preparation from the ear showing discrete colonies of *A. fumigatus* mycelium 10 per cent. KOH × 514.

of yeast, or even of mould, which when present could be detected amongst these bacteria on microscopical examination.

Microscopical Methods

Moulds. When the position of the mould growth in the affected external auditory meatus was ascertained a sterile closed bacteriological wire loop was inserted into the meatus and some of the material withdrawn from it,

placed in a drop of 10 per cent. caustic potash solution on a microscopical slide, and covered with a coverslip. This preparation was warmed slightly over a spirit lamp flame, and then examined microscopically, using a binocular microscope with 8× oculars and the 2/3 and 1/6 objectives. If

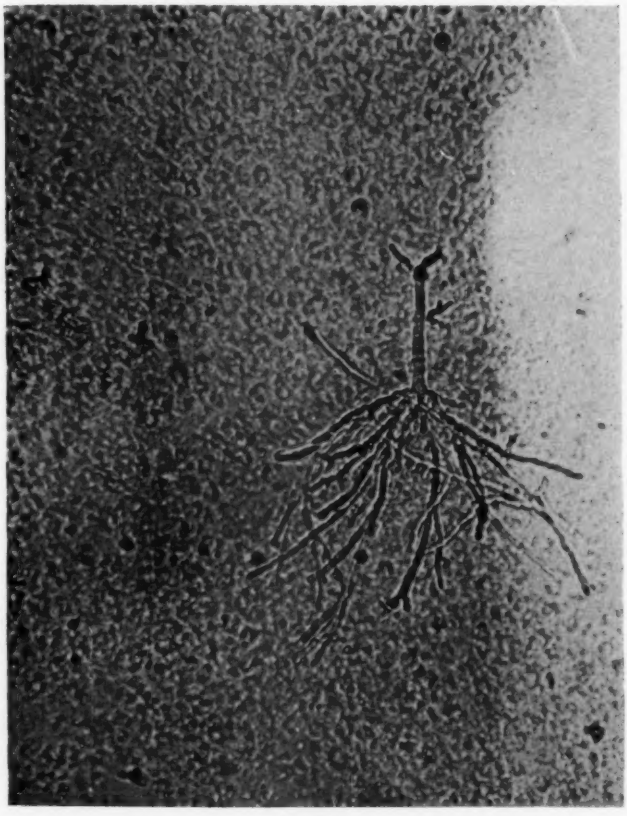


FIG. 4*b*.
Direct preparation from the ear showing minute root-like structures formed by mycelium of *A. niger*, 10 per cent. KOH × 200.

conidiophores had already been observed during the superficial examination it was usually possible to see them microscopically also. Sometimes these conidiophores were sufficiently well developed and characteristic to permit of species identification; at other times it was necessary to await the results of culture before a definite specific identification could be made. When only vegetative mycelium was present in the preparation this, in

most cases, had not already been seen on superficial examination. In such cases, since the mycelium is not usually characteristic of a species, the only means of identification was by culture of the organism whereby

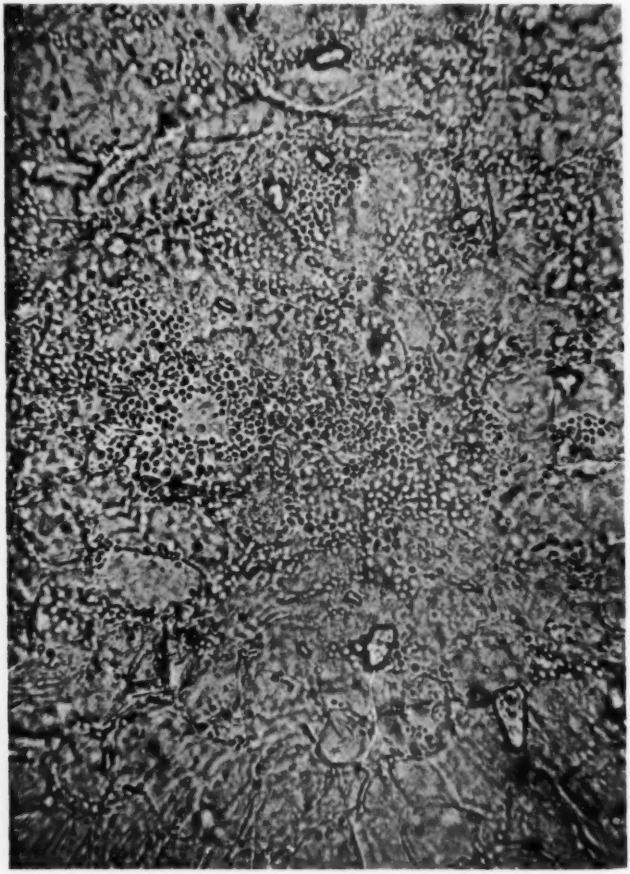


FIG. 5*a*.

Direct preparation from the ear showing budding cells of *C. albicans*, 10 per cent. KOH × 514.

the reproductive structures could be studied. Certain features, however, were present in the vegetative structures of some species so frequently that it was tempting to forecast the species when they were observed, and it is possible that, with increasing familiarity, these features might become of some significance in diagnosis. Thus, in the case of *A. fumigatus*

in otomycosis lesions, the hyphae were nearly always relatively thick and frequently blunt and forked at the tip; occasionally discrete little colonies of this type were seen (Fig. 4*a*). Again, in the case of *A. niger*

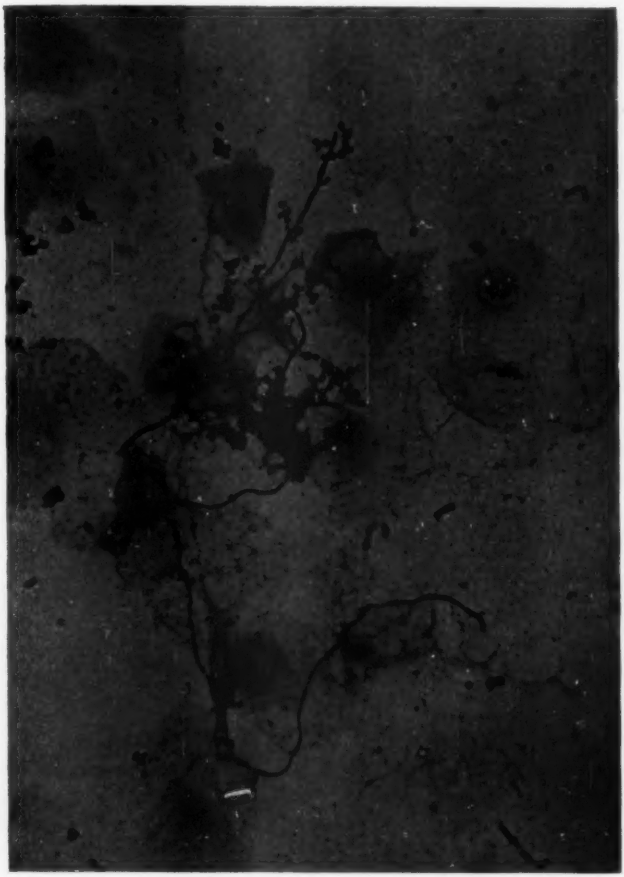


FIG. 5*b*.

Direct preparation from the ear showing mycelium and budding cells of *C. albicans*, Gram stain × 514.

a recurrent feature was a minute root-like system (Fig. 4*b*). Such vegetative growths are not seen in the mycelium of these species on standard culture media.

In addition to mounting material in 10 per cent. caustic potash solution it was sometimes desirable to mount some of it in lactophenol

and cotton blue (Amann's medium) so that it could be preserved for future reference. This medium contains: lactic acid, 20 parts; phenol, 20 parts; glycerol, 40 parts; and distilled water 20 parts. Cotton blue dye is added as ·05 per cent. by weight. In some cases also smears of the material were fixed, and stained variously, by Gram, P.A.S. or Grocott's modification of Gomori's silver methenamine impregnation method.

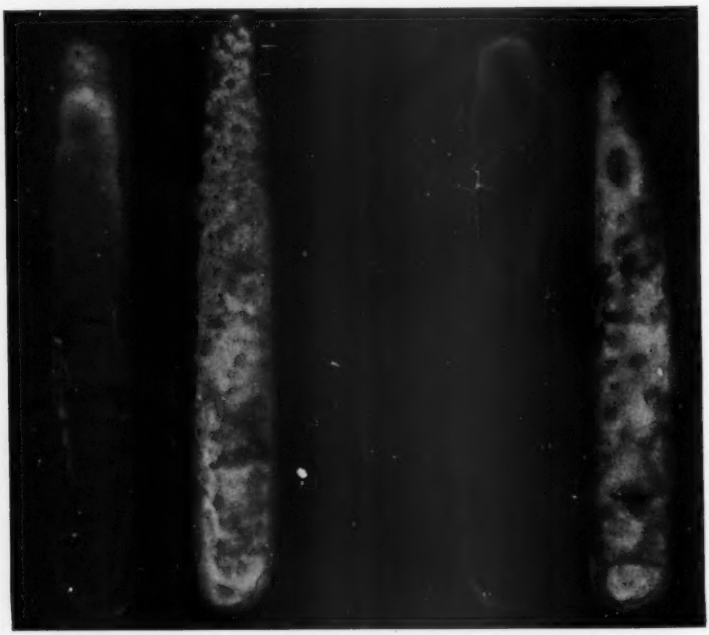


FIG. 6.

Comparison of sporulation at two different incubation temperatures after 48 hours culture. (Sporulation indicated by darkening of the surface of the culture).

Microscopical Examination of Material Containing Yeast-like Fungi.

Material suspected of containing yeast-like fungi was also mounted in 10 per cent. caustic potash solution as in the case of material suspected of containing mould. Smears were also prepared for staining by Gram or sometimes by P.A.S. These yeast-like fungi could be detected in the caustic potash solution as small ovoid budding and highly refractive cells (Fig. 5*a*). Occasionally they were associated with the pseudomycelium or true mycelium which is a different growth phase of the same organism (Fig. 5*b*). They were not always revealed by Gram stain and this could not therefore be relied upon as the sole test for their presence or absence.

Culture of Material from Affected Ears

Material removed from the affected ears was cultured on Sabouraud glucose agar slopes in most cases. In some cases, honey-peptone agar slopes were used (the peptone incorporated in this medium was "Oxoid" bacteriological peptone). An antibiotic mixture of penicillin and streptomycin was included in the medium in such quantity as to give a final concentration of 20 units of penicillin and 40 units of streptomycin per ml., but more recently chloromycetin has been used since it can be autoclaved with the medium and is equally as effective in suppressing bacterial growth in the cultures. It is incorporated as 0·5 mg. per ml. Cultures were incubated at 37° C. in order to confirm that the fungus observed microscopically could grow at the temperature prevailing in the external auditory meatus. They were cultured at 27° C. also in case the mould was a mere contaminant, and, more important, in order to prevent the overgrowth of yeast-like organisms by fast-growing moulds such as *A. fumigatus* and *A. niger* which could well happen in cultures incubated at the higher temperature.

All the species of Aspergillus isolated in this series grew and sporulated rapidly at 37° C., while at 27° C. growth and sporulation proceeded at a slower rate. In the case of primary cultures of *A. fumigatus* incubated at 37° C. growth was generally visible to the naked eye within 24 hours, by which time sporulation also had commenced. This was often the case with *A. niger* also. Other species incubated at this temperature often produced growth also within this period. In sub-culture at this temperature growth and sporulation were even more rapid (Fig. 6).

Superficial Cultural Features of the Species Isolated from Patients' Ears.

A. fumigatus. Primary cultures incubated at 37° C. usually showed growth visible to the naked eye within 24 hours. Sporulation was indicated by a pale blue colour which spread gradually over an otherwise white superficial growth; this colour deepened and changed to a greyish blue-green with age.

A. niger. Primary cultures incubated at 37° C. produced white superficial growth visible to the naked eye usually within 24 hours. This became gradually lemon yellow in patches at the onset of sporulation and then changed gradually to dark brown when the conidia were mature.

A. terreus. Primary cultures at 37° C. revealed superficial growth usually within 24 hours but sometimes took longer to do so. This again was white at first, but became gradually tinged with a light buff colour as sporulation proceeded, and became a deeper colour as the conidia matured.

A. nidulans. Primary cultures at 37° C. produced white superficial growth visible to the naked eye, usually within 24 hours, but sometimes took longer. The onset of sporulation was indicated by the

appearance of a pale green colour which deepened in due course. In addition to the conidiophores which were represented by this green colour, special sporing structures (perithecia) developed within a few days amongst the conidial mass. They were spherical, measuring 1-2 mm. in diameter and were ochraceous to salmon in colour.

A. flavus. Primary cultures at 37° C. produced white growth visible to the naked eye, usually within 24 hours. This changed gradually to greenish yellow as sporulation proceeded and then in due course to lime green. Some isolates produced, in addition to the conidiophores, greyish, spherical structures measuring about 1-2 mm. called sclerotia which differ from perithecia in so far as they are sterile.

A. candidus. Primary cultures produced white superficial growth within 24 hours or shortly after, at 37° C. This became more dense and white as conidiophores developed and sporulated. Older cultures did not change colour or at most, became cream-coloured. On the other hand, in these strains, the reverse was at first ochraceous, then deep red, and finally black.

A. glaucus. Primary cultures were at first white at 37° C., becoming pale green or blue-green as conidia developed. This gradually darkened and became in due course brown. In addition to the conidiophores, yellow perithecia measuring about 0·5 mm. to 1 mm. in diameter were produced.

Microscopical Features of Aspergillus spp.

General. In all species the conidiophores, which provide the chief criteria for identification purposes, are aerial, erect, usually unbranched stalks borne on the aerial mycelium. They are swollen at the tip to form the vesicle. This part of the conidiophore varies somewhat in shape according to the species and even to some extent in strains of the same species. It gives rise to a large number of short, more or less closely packed stalks which develop the conidia successively at their tips so that long chains of conidia are produced, or else, as in a number of species, form a second rank of short stalks which bear the conidia. When there are thus two ranks of short stalks, one upon the other, those of the first or proximal rank are known as metulae, while those of the second or distal rank are known as phialides. In some species the conidia form radiating chains, giving the conidial head a spherical appearance when viewed with a lens; while in other species they form chains in columns or plumes. According to the species the conidia vary in colour which usually changes with age, and are spherical or elliptical or sometimes barrel-shaped, and finally may be rough or smooth according to species. In species which produce perithecia, the spores contained within them (ascospores) are at first enclosed in fours in minute transparent sacs (asci). These ascospores are variously coloured according to the species.

Main Microscopical Features of Species in Culture on Sabouraud Glucose Agar Medium

A. fumigatus. The conidiophores vary in length and width, even in the same culture. Typically, the proximal part of the vesicle merges very gradually into the stalk. This is due to the upper part of the stalk being almost as wide as the vesicle which surmounts it. The phialides are borne typically on the upper half of the vesicle, lying in the axis of the conidiophore, and are packed side by side in a palisade-like manner

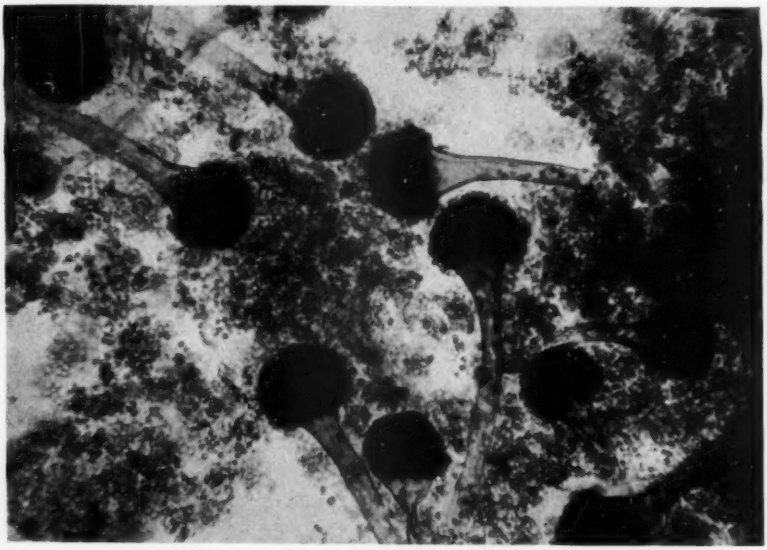


FIG. 7.

Direct preparation from the ear showing conidiophores of *A. fumigatus* with palisade-like arrangement of phialides. Lacto-phenol and Cotton blue × 514.

(Fig. 7). In some strains the phialides may occupy a greater part of the vesicle surface and in others both types of arrangement of these phialides may be encountered in the same culture. The conidia are globose and finely roughened. They measure, according to Thom and Raper (1945) 2-3·5 μ in diameter.

A. niger. The conidiophores are long, relatively broad, erect and thick-walled. They are surmounted by a large spherical vesicle from all parts of which radiate metulae which in turn bear phialides. The conidia vary from chocolate brown to purplish brown and are globose and rough-walled. They measure, according to Thom and Raper 2·5-4 μ in diameter. The outline of the conidial head is spherical when undisturbed (Fig. 8).

A. terreus. The conidiophores are relatively long and are surmounted by a globose or subglobose vesicle bearing metulae and phialides. The conidia are light brown, globose and are smooth-walled. They measure,

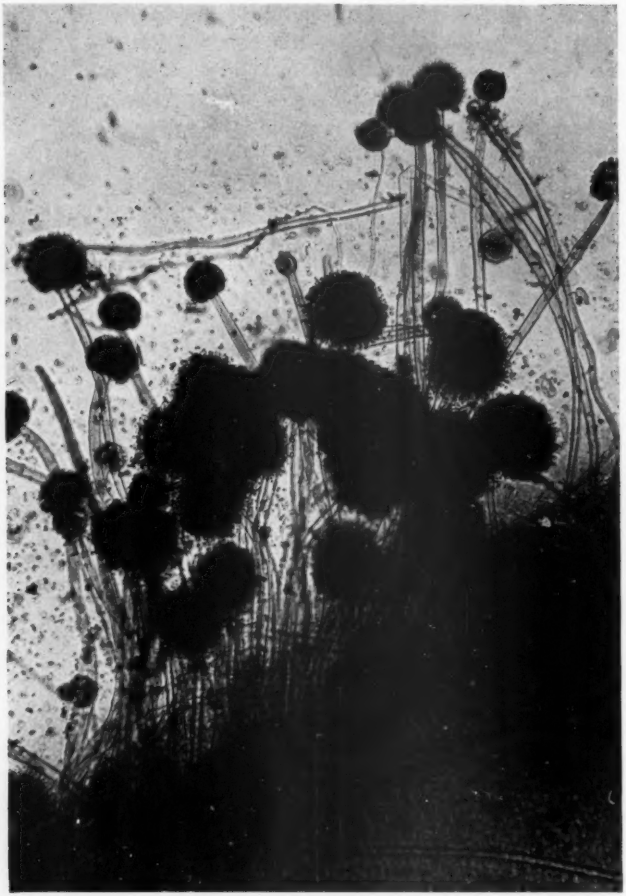


FIG. 8.

Direct preparation from the ear showing conidiophores of *A. niger* in different stages of maturity. Lacto-phenol and Cotton blue × 120.

according to Thom and Raper 1·8-2·4 μ in diameter. At maturity, they are, when undisturbed, closely packed in columns. A special and characteristic feature of the vegetative mycelium is the development of large numbers of spherical, short-stalked spores along the hyphae. In time these enlarge and

become thick-walled, and then closely resemble the Hülle cells seen in association with the perithecia in *A. nidulans*. This feature has been observed also by Austwick (1959).

FIG. 9.
Direct preparation from the ear showing conidiophores of *A. nidulans*. Lacto-phenol and Cotton blue ×514.

A. flavus. The conidiophores in this species are relatively long. The vesicle is globose or subglobose, and the stalk is roughened owing to the presence of granules on the wall, or, in some cases, pits. The vesicles bear phialides only, or both metulae and phialides. The conidia measure,

according to Thom and Raper, between 3 and 5μ. At maturity they form loose columns.

A. nidulans. The conidiophores are relatively short (Figs. 9 and 10*a*). They are surmounted by a distinct subglobose vesicle bearing both metulae and phialides. The stalks are coloured characteristically a light reddish brown. The conidia are globose, finely rough-walled, and measure, according to Thom and Raper 3-3·5 μ in diameter. At maturity the undisturbed conidial head is columnar. The perithecia contain dark reddish or purple ascospores. They are surrounded by numerous hyaline thick-walled cells called Hülle cells (Fig. 10*b*).

A. candidus. The conidiophores show much variation in size and structure, including the detailed structure of the metulae. The conidia of strains seen in this series were globose to subglobose and smooth-walled. According to Thom and Raper they may be elliptical or barrel-shaped in some strains and measure 2·5-3·5 μ.

Cultural Features of Yeast-like Fungi

Material suspected of containing "yeast" was cultured at 37° C. on the same media as those used for species of Aspergillus. Yeast-like colonies were usually visible within 24 hours to the naked eye. They were at first circumscribed and pellucid but in due course became more irregular in outline and opaque milk white to creamy white. Identification was carried out on potato-carrot extract agar containing bile and made up according to the formula recommended by Segretain *et al.* (1958) as well as by fermentation tests when the species was other than *C. albicans*.

Identification of C. albicans on Potato-Carrot Extract Agar plus Bile

Yeast from primary colonies was streaked in crossed parallel lines on to the surface of the potato-carrot agar medium in Petri dishes and then dragged into the medium along these lines with the point of a sterile mounted needle. The Petri dish cultures were then incubated at 20° C. Within 24 hours or sometimes after a longer period, mycelium grew out from the sides of the streaks of yeast-cells. Sooner or later, in the case of *C. albicans*, large sperical, thick-walled cells (Chlamydospores) were produced at the tips of short branches or at the tips of the main hyphae.

Treatment

Since one of the problems to be considered in this investigation was the significance of fungus invasion in the perpetuation of otorrhœa, it was desirable that therapy should be directed solely at the fungus and should have no significant effect on bacteria.

It was decided, therefore, to use "Nystatin" (Squibb & Co.), one of a series of substances developed for their antifungal properties, parti-

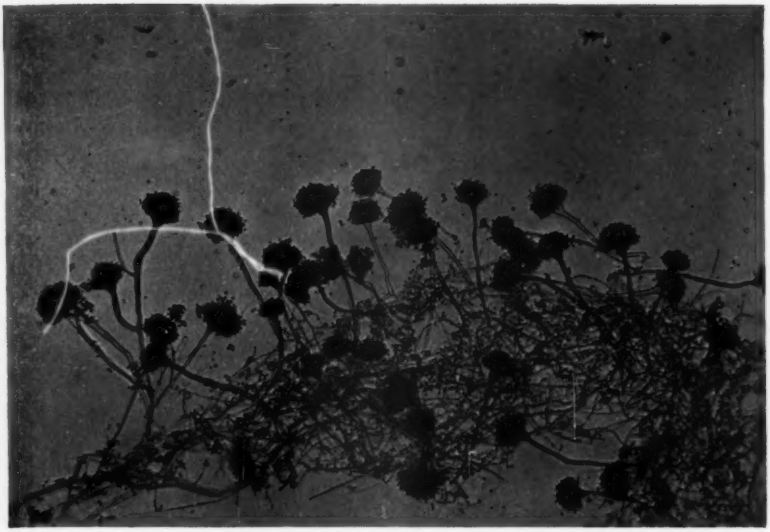


FIG. 10*a*.
Conidiophores of *A. nidulans* from culture. Lacto-phenol and Cotton blue × 200.

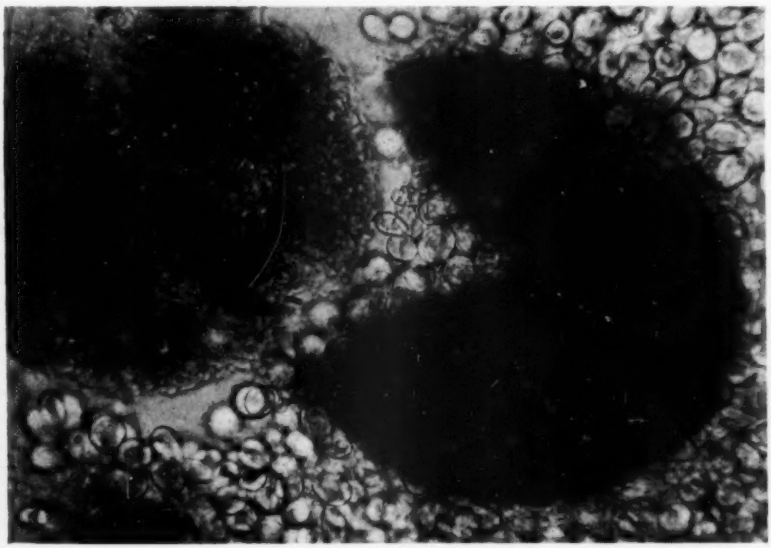


FIG. 10*b*.
Ruptured perithecium of *A. nidulans* from culture, and extruded mass including ascospores. Numerous thick-walled Hülle cells surrounding perithecium. Lacto-phenol and Cotton blue × 514.

cularly against yeasts. The substance was said to have no significant effect on bacteria, and there had been no previous assessment of its value in otomycosis.

A routine was evolved where out-patient treatment was given for a period of three weeks. It was thought that such a period would cover adequately all the phases in the life of the invading fungi. This was necessary for there was no information about the phase in which the fungus was most susceptible to Nystatin, and it was known that antibiotics are usually only effective against bacteria in some particular phase of their life-cycle.

Since most fungi require some moisture for growth, it was decided to use a form of treatment which would eliminate moisture as far as possible. Dry mopping of the meatus formed an essential part of this, and it was obviously illogical to use any aqueous vehicle for the Nystatin. For the majority of the cases, Nystatin was applied in a powder of which boric powder formed the base (100,000 units of Nystatin per gramme of powder). Recently lactose has been used as the base without any reduction in efficiency.

The routine thus evolved consisted of attendance by the patient at the out-patient treatment clinic three times weekly for three weeks. At each attendance the meatus or radical mastoidectomy cavity was meticulously cleansed by dry mopping and the manual removal of any large pieces of debris. The epithelium of the meatus or cavity was then lightly "frosted" with the powder containing Nystatin. The patient was accepted as cured when the meatus showed a little secretion of normal wax.

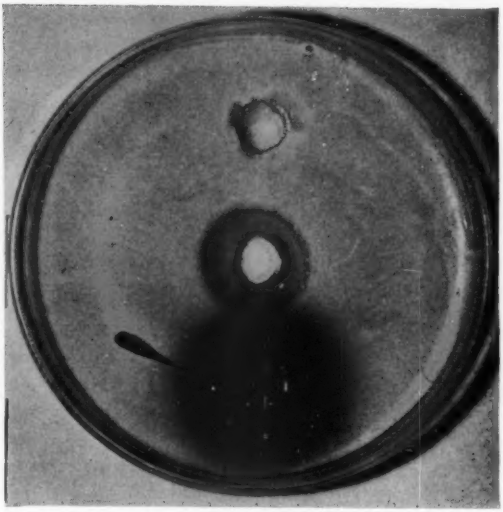
Results

Of the 83 cases diagnosed as fungus infection, 2 were not prepared to attend for treatment, and 7 cannot be traced. A further 7 required more prolonged therapy or relapsed soon after they were thought to be cured. These relapses responded to further periods of therapy with Nystatin.

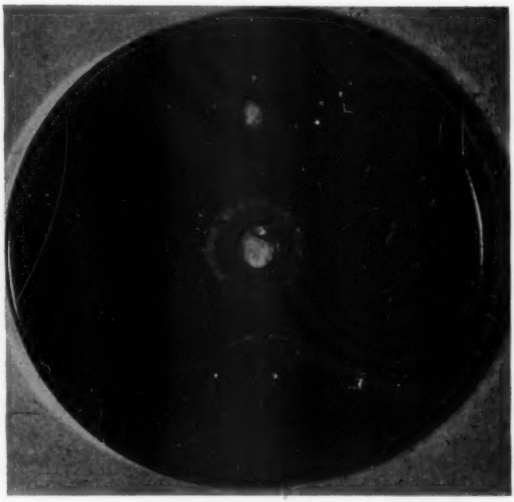
The remaining patents remained free of symptoms.

The period of therapy was uneventful in the main, but in 10 patients it was complicated by the development of an aural furunculosis. This may have been due to an associated staphylococcal infection which the fungal metabolites, being partly of the nature of antibiotics, had been holding partly in check, only to come to light with the elimination of the fungus. The development of furunculosis may, therefore, be a useful guide to the elimination of the fungus.

Unfortunately an initial bacteriological assessment was not done in all these cases. Of the 5 patients who had *Staphylococcus aureus* at an initial investigation, only one developed a furuncle while under treatment with Nystatin. It is possible, therefore, that cross-infection by the "hospital staphylococcus" may have occurred during treatment.



C. albicans



A. niger

FIG. 11.

Comparison of the inhibiting effect of fungistatic and fungicidal agents upon the growth of *C. albicans* and *A. niger* in culture.

Although many substances appear to be efficient fungicides, Nystatin has shown two properties not possessed by most of them. Its application does not cause pain, nor does it colour the meatus. Many of the substances used in the past could cause intense pain—tincture of iodine, thymol and metacresyl acetate (Whalen, 1938)—and it was not unknown for patients to default from treatment, possibly because they found it more intolerable than the disease. The aniline dyes—gentian violet, brilliant green—have been proved most efficient fungicides *in vitro*, but they stain the meatus to such an extent that cure must be well-nigh impossible to assess.

Some authors in the past have dismissed the matter too lightly by claiming that Industrial Spirit used locally inhibits all fungi. This may be true of concentrations greater than 70 per cent. but such concentrations are also lethal to epithelial cells. The standard B.N.F. concentration of 50 per cent. is singularly ineffective, at least *in vitro* (Fig. 11).

Conclusion

It is not claimed that this search for fungus infection has solved the otologist's perennial problem of the discharging ear, for rather more than 50 per cent. of the patients investigated gave no evidence of a growing fungus. Nevertheless, this does seem to be a field worthy of further investigation, and the writers are only too well aware of the many unsolved problems which remain. The control of such investigations may be hampered for some time by the unavailability of mycologists in hospital practice.

From the clinical aspect it cannot be stressed too much that diagnosis is still inaccurate, and the clinician must rely upon the control of his mycologist colleague. With such control, however, the new fungicidal agents can provide a valuable addition to the otologist's armamentarium.

Summary

1. During the years 1956-9, 180 patients with intractable otorrhœa, due to either otitis externa or infection of radical mastoidectomy and fenestration cavities, were investigated for the possibility of an active fungus infection.

2. Eighty-three of these patients had positive evidence of such an infection, and were treated with a specific fungicidal agent with resolution of the otorrhœa.

3. The clinical picture of fungus infection in the external ear is described, but the inaccuracy of clinical diagnosis is emphasized, and criteria for diagnosis are given.

4. The microscopic and cultural characteristics of the fungi most commonly found are described.

5. The ætiology of fungus infection of the external ear is discussed briefly. Knowledge of this aspect of the condition is scanty.

6. Possible methods of therapy are discussed, and the value of new fungicidal agents is considered.

Acknowledgments

We are indebted to Mr. T. McM. Boyle, Mr. O. C. Lord, Mr. C. P. Mills, and the late Mr. G. S. Seed for permission to carry out this investigation on patients under their care.

We would like to recognize the help that we have received from Mr. A. L. Pegg, of the University of Leeds Department of Medical Photography, in obtaining the illustrations of the clinical condition, and it is regretted that cost prevented the reproduction of these plates in their original colours. We would like to thank Mr. R. A. Forster of the Mycology Section, Department of Dermatology, United Leeds Hospitals for his help in the preparation of cultures.

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"Claremont", Bramham Road,
Thorner, Near Leeds

AN APPROACH TO THE PITUITARY *VIÂ* A NASAL OSTEOPLASTIC FLAP

By R. G. MACBETH (Oxford)*

THE object of this communication is to describe what is believed to be a new trans-nasal approach to the pituitary. In consequence no attempt will be made to discuss the scientific bases of hypophysectomy nor to assess results.

In the past the main object of approaching the pituitary was to deal with tumours in this region but latterly a normal gland has become of surgical interest because of the need to remove it in certain cases of advanced carcinoma of the breast and prostate.

Other Routes *viâ* the Nasal Apparatus

All these routes ultimately lead to the sphenoidal sinuses and thence to the pituitary fossa. Hamberger has had a wide experience of removal of the pituitary in Sweden, and Bateman has been the pioneer in this country.

(*a*) *Trans-septal.* This has hitherto been associated with the name of Cushing, and may be achieved from the nose or from the mouth. Its main application has been in cystic tumours of the pituitary where a relatively narrow field is less of a disadvantage, the object being to suck out the tumour. The distance from the surface and the narrow field, is, however, a grave disadvantage when removal of a gland is attempted.

(*b*) *Trans-palatal.* This is a comparatively short route, but the approach is through an infected field and it is not easy to bring the microscope to bear through the mouth.

(*c*) *Trans-ethmoidal.* This route was originally described by Chiari and was advocated by Nager in his Semon Lecture in 1939. It has been used with conspicuous success by Bateman. Although it gives a comparatively wide field of approach and short depth of wound and although it can be combined with a septal resection, the obliquity of access still remains and this may make the recognition of the pituitary fossa less easy than if one can view the area from the midline.

(*d*) *Trans-antro-ethmoidal.* This route has been extensively used by Hamberger, provides a wide deep entry wound, but the application of the microscope is inconvenient.

* From the Department of Otolaryngology, United Oxford Hospitals.

FIG. 1.

FIG. 2.

The Osteoplastic Operation

Pre-operative Preparation. The patient is given whatever premedication the anæsthetist fancies, but the nasal cavity should be painted up with cocaine paste 25 per cent., and the whole nasal bridge and the face over the ethmoidal and upper maxilla are infiltrated subcutaneously with xylocaine ½ per cent. containing adrenalin 1:100,000. Penicillin and Sulphadiazine "cover" is commenced, and the patient is given hydrocortisone 100 mg.

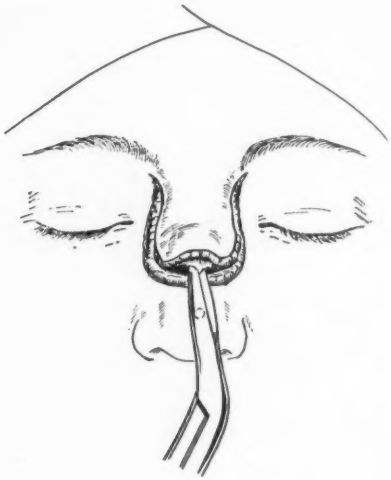

FIG. 3.

Anæsthesia. An endotracheal general anæsthetic should be given unless the anæsthetist thinks the patient's condition contraindicates this, in which case the local anæsthetic mentioned above reinforced with appropriate nerve blocks could be employed.

The Operation

(1) The incision is as for a bilateral external ethmoidectomy the lower ends of the two limbs being joined by a curved incision, which crosses the nose just below the junction of the nasal bones and the lateral nasal cartilages (Fig. 1).

(2) The incision is deepened to bone laterally; over the bridge the nasal cartilages are severed from the nasal bones and the nasal cavities are entered (Fig. 2).

(3) The mucous membrane of the lateral nasal wall is freed from

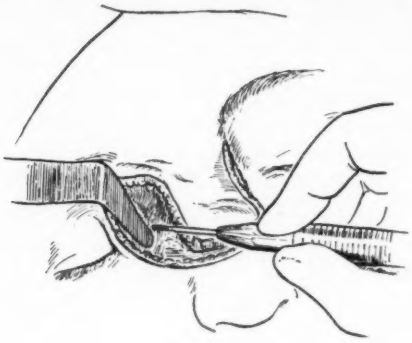

FIG. 4.

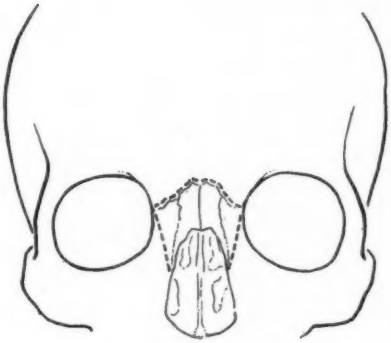

FIG. 5.

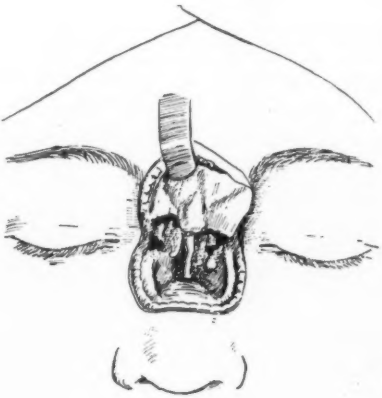

FIG. 6.

the bone by blunt dissection and the septal cartilage is freed by means of scissors from the nasal bones superiorly (Fig. 3).

(4) The lacrimal sacs are retracted laterally on each side and the nasal processes of the maxillae are cut across by means of a cylindrical dental burr. The bony incision commences at the point where the fronto-maxillary suture joins the orbital margin and it runs to about half-way down the lateral nasal wall (Fig. 4). Irrigation and suction are used throughout this part of the operation.

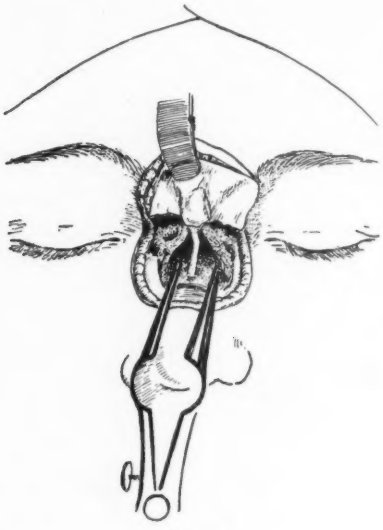

FIG. 7.

(5) These portions of the maxillae together with the nasal bones are now freed from the frontal bones by a sharp tap along the suture lines, using a small osteotome and hammer. The area of the bone flap so reflected is shown in Fig. 5.

(6) The osteoplastic flap is turned upwards and held out of the way by means of a retractor (Fig. 6).

(7) A submucous resection of the septum is now easily and rapidly carried out from above (Fig. 7) and the alae of the vomer are exposed and removed. In doing this the mucosal flaps are detached and torn away from the sphenoid.

(8) The removal of the vomerine alae opens the sphenoidal sinuses. At this point the operation on the pituitary differs only in detail from the routine adopted when other routes are used. The sphenoidal sinuses have been as widely opened as seems needful and extra room may be gained

if needed by opening the ethmoid. The operating microscope is now brought into action, with its directional axis strictly in the midline. This makes the recognition of the bulge of the pituitary fossa into the sphenoidal sinuses relatively easy, particularly when the field is compared with the skiagrams taken before operation. If there is any doubt about

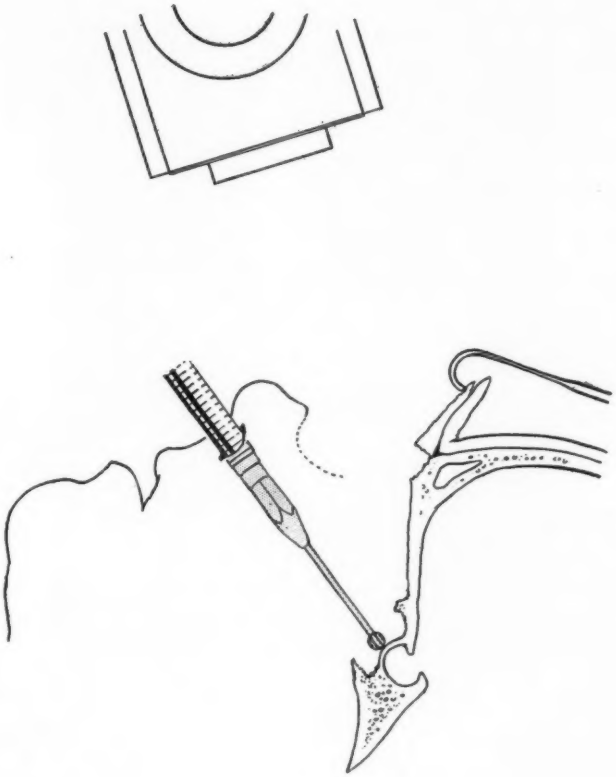

FIG. 8.

the position of the floor of the pituitary fossa, check radiographs may be taken on the table, but these are usually unnecessary when this approach is used.

A convenient method of opening the fossa is by means of a small dental burr and the drill handle which carries it may be passed *viâ* one or other nostril (Fig. 8).

The incision of the dural layers and removal of the gland follows the

predilection of the operator. I favour a cruciate incision near the back of the fossa through both dural layers and extraction of the gland piecemeal from within its pseudo-capsule. A muscle graft may be needed in order to stop the inevitable flow of cerebrospinal fluid and a pack may be required. The nostrils are packed as for any septum operation at the surgeon's discretion.

FIG. 9.

(9) The osteoplastic flap is now replaced and the soft tissue layers and skin are carefully sutured.

(10) The final stage is to deal with the patient's "broken nose". For this I use strips of ribbon gauze soaked in collodion and place a piece of Ash's soft metal between the layers.

Post-operative Treatment

The patient is given hydrocortisone 100 mg. 2 hours after operation and a maintenance dose of 50 mg. 4 times a day thereafter. She may require small doses of thyroid. The packing in the nostrils is removed in accordance with the surgeon's routine, and the pack which goes to the

pituitary fossa can usually be removed in 48 hours. The nasal splintage remains in position for a week to ten days, and the stitches are removed when it comes off. (Since these patients are often in very poor general condition it is a counsel of wisdom for the rhinologist to arrange disposal of the patient before admitting her to the E.N.T. Ward.)

Discussion

I have used this operative approach upon 7 patients. It provides a midline and relatively short route to the pituitary fossa, and these are its main advantages. In addition the operative exploration involves a minimum of nasal sinus surgery. This may be an advantage where infection may be suspected, or where it cannot be entirely ruled out.

Healing of the skin incision is rapid and the scarring is minimal (see Fig. 9).

Although the making of the osteoplastic flap and its replacement adds something to the length of time needed, the whole operation can usually be carried out within 2 hours. Incidentally it may be added that this particular approach has been found useful for the exploration of the sphenoidal sinuses in the case of a tumour arising in that situation.

The main disadvantage is that shared by all trans-nasal approaches to the pituitary—namely the escape of cerebrospinal fluid and the danger of meningitis. An adequate muscle-graft locally applied to the pituitary fossa, together with anti-bacterial therapy would appear to counteract this danger.

Acknowledgments

I must record my grateful thanks to Professor Hamberger and Mr. Geoffrey Bateman, who kindly allowed me to see their techniques and discussed them with me. Without a knowledge of these I could not have devised the above method.

Additionally I wish to thank Miss A. Arnott and Miss M. McLarty, the Hospital artists, for the illustrations.

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The United Oxford Hospitals,
Department of Otolaryngology
The Radcliffe Infirmary,
Oxford

CLINICAL RECORDS

INTRAMURAL ŒSOPHAGEAL FOREIGN BODY

By W. LEACH (London)

THE passage of a foreign body through the wall of the gullet into the soft tissues is a relatively uncommon occurrence. It is even less common to discover one embedded in the wall of the pharynx or œsophagus. There are few reports of methods of dealing with such emergencies over the past ten years or so. The case to be described is the third perforating foreign body seen at St. Thomas' Hospital in the past five years. The first was reported by Bateman and Carruthers (1956) and the second is reported elsewhere in this issue. This third case presented within three months of the previous one.

Case Report

The patient was a West Indian woman aged twenty-six, who had swallowed a meat bone the day before she presented herself in the Casualty Department. She had tried to encourage the bone to pass on by swallowing bread and butter. Her pain on swallowing gradually became worse over the next twenty-four hours and by the time she was seen it was constant. She was tender just to the right of the cricoid cartilage, but otherwise there were no abnormal physical signs and indirect laryngoscopy revealed no abnormality. The presence of a small linear foreign body was confirmed radiologically and it appeared to be lying in the œsophagus just below the cricopharyngeal sphincter (Fig. 1). However, œsophagoscopy revealed nothing more than a vertical linear tear of the mucosa and a streak of blood about 1·5 cm. below the cricopharyngeal sphincter on the posterior wall of the œsophagus.

The patient was started on antibiotics and given water only overnight. An X-ray next morning showed the foreign body in almost the same position as previously with a little increase in the swelling of the post-cricoid soft tissue. A further œsophagoscopy was performed, at which a pair of Paterson's forceps was carefully passed through the mucosal tear but no foreign body could be felt. Accordingly the neck was explored without delay using an incision along the anterior border of the right sternomastoid muscle. The omohyoid muscle was divided and the sternomastoid and carotid sheath were retracted laterally. The lateral lobe of the thyroid gland was displaced forwards and medially to reveal the œsophagus. An extensive search revealed no sign of a foreign body. A bronchoscope was passed into the œsophagus just beyond the cricopharyngeal sphincter. Using the distal illumination from this instrument it was hoped to show up the foreign body by trans-illumination through the œsophageal wall. This, however, was of no avail, nor could any hard object be felt by palpation

of the œsophagus against the metal bronchoscope. A small vertical incision was then made in the muscle layer of the postero-lateral aspect of the œsophagus just below the sphincter exactly where the foreign body was thought to lie. A further search between the muscle and mucosal layers failed to reveal the foreign body; it was therefore decided to place two Michel clips as markers, one at the upper and one at the lower end of the œsophageal incision. This done,

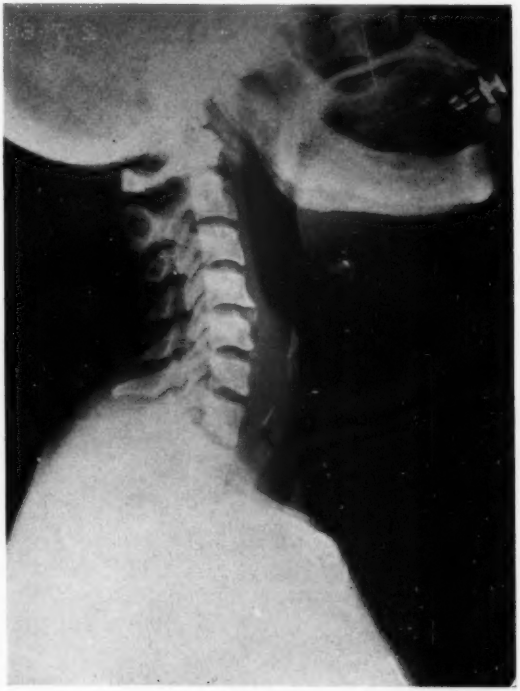

FIG. 1.
(*a*) Direct lateral.

the incision was repaired with catgut and the wound packed with ribbon gauze soaked in penicillin and sulphathiazole solution. A feeding tube was passed through the nose into the stomach, the patient returned to the ward and antibiotics continued.

Next morning, further X-rays taken in several different planes showed the lower tip of the foreign body to be only about 5 mm. above and very slightly to the left of the upper marker (Fig. 2). A second exploration was now embarked upon and the original wound reopened. The foreign body was eventually found by palpation against a bronchoscope in the œsophagus, but only after the muscle layer of the œsophagus had been opened up again wide enough to allow a finger to be passed through the incision. A sharply pointed sliver of bone 2·4 cm. long

(Fig. 3) was removed from between the mucosa and muscle layer of the right postero-lateral wall of the œsophagus where it had lain obliquely 1 cm. below the sphincter, having pierced and traversed the mucous membrane through the tear already seen. The œsophagus was repaired in two layers and the wound closed with drainage for forty-eight hours. Tube feeding was continued for six days, followed by a liquid diet for three days, after which the patient was able

FIG. 1.

(*b*) Left oblique view in which the foreign body appears lying across the tip of a small cervical rib.

X-rays taken prior to œsophagoscopy showing the foreign body (indicated by arrows) and swelling of the surrounding post-cricoid soft tissues.

to swallow soft food without pain. She left hospital exactly two weeks after she had swallowed the bone, and indirect laryngoscopy revealed no abnormality when she attended for follow-up in a further fortnight.

Discussion

The problem of dealing with perforating foreign bodies has been somewhat simplified since the advent of modern anæsthetic and surgical techniques, and in particular antibiotics. Prior to 1940 there are few reports of perforating

FIG. 2.

X-ray taken after the first exploration with feeding tube and markers in position. The foreign body has moved slightly and is lying with its lower tip very close to the upper marker.

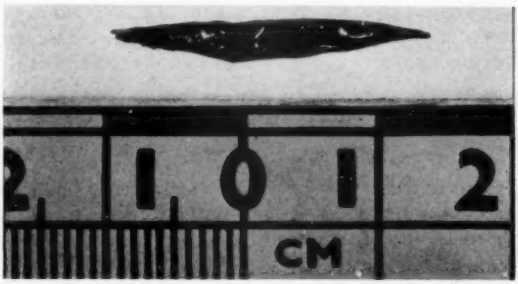

FIG. 3.

The foreign body—a sharp-pointed sliver of meat bone—after extraction.

foreign bodies being successfully located and removed, and presumably surgeons were naturally reluctant to explore a potentially infected field after œsophagoscopy had failed. There are reports of perforating foreign bodies being removed endoscopically, but many were left so that abscess formation or mediastinitis and often death supervened. The surgery of their removal was thus largely the therapeutic surgery of drainage of the resultant cervical abscess. In the last ten years or so there are reports of more determined efforts to remove perforating and intramural foreign bodies as an emergency prophylactic procedure. Arnold and Som (1954) described a similar technique using markers to locate and extract a foreign body embedded in the posterior pharyngeal wall *viâ* an internal approach.

The use of an endoscope as advocated by Bateman and Carruthers (1956) materially assists in the location of foreign bodies not so much as a transilluminator but as a hard surface against which the œsophageal wall can be palpated. In this present case palpation against the firm rubber feeding tube proved useless and it was not until a bronchoscope had been passed during the second exploration that the foreign body could be felt. Markers were used, as in the case reported by Hammond (*vide* supra) and the operative technique was closely similar in these two cases.

It seems incredible that after good radiological localization these intramural foreign bodies should prove so difficult to find on exploration. This raises the question of whether a one-stage procedure should be planned for every case of a suspected perforating foreign body, that is all those in which the lumen of the gullet has been first shown by endoscopy to be perforated and confirmation of a foreign body in the neck has been obtained subsequently by X-ray. Using a one-stage procedure, the two explorations could be combined into one single operation with a short break in the middle for taking X-rays in the theatre and perusal of the wet films showing markers in position.

Summary and Conclusion

A report is given of a third case from the same hospital where a foreign body had perforated the gullet and lodged in the soft tissues, in this instance the œsophageal wall. The two previous reports are quoted and the technique of exploration was similar for all three cases, involving the use of an endoscope and markers where necessary. The complexity of the problem of how to manage these perforating foreign bodies as surgical emergencies is discussed in the light of modern knowledge and a suggestion made as to a possible future method of approach using a planned one-stage technique.

Acknowledgments

I am grateful to Mr. G. H. Bateman for permission to publish details of this case and for his advice and guidance in preparing the paper. I should like to acknowledge Mr. W. J. D. Bradfield's kindness in referring the patient and his assistance at each of the operations. My thanks are also due to the X-ray Department of St Thomas' Hospital for the loan of X-ray films and to the Department of Clinical Photography for supplying me with the photographs.

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E.N.T. Department,
St. Thomas' Hospital,
London, S.E.1

PERFORATING FOREIGN BODY OF PHARYNX

By V. T. HAMMOND (London)

Sharp foreign bodies in the pharynx are a fairly common occurrence and are usually fish or meat bones. However, the passage of the foreign body through the pharyngeal wall into the soft tissues of the neck fortunately occurs very infrequently. This case is of interest both in the unusual type of foreign body and the position in which it came to rest.

Mr. W. M., aged 54, was seen in the Casualty Department at St. Thomas' Hospital on *April 5th, 1960*, complaining of pain in the left side of the throat on swallowing. The pain had come on while he was eating tinned pilchards for lunch.

On examination he was a little tender on the left side of the neck at the level of the cricoid cartilage. No abnormality was seen on indirect laryngoscopy. A lateral X-ray of the neck revealed a linear foreign body lying horizontally at the level of the upper border of the cricoid cartilage (Fig. 1).

He was admitted to hospital and an œsophagoscopy was performed. There was a small puncture wound in the lateral wall of the left pyriform fossa but no foreign body was visible. Gentle probing of the puncture wound failed to reveal the foreign body.

On recovering from the general anæsthetic the patient was re-X-rayed. The foreign body had not altered in position but there was some increase in the width of the post-cricoid shadow as compared with the previous X-rays. (Fig. 2).

It was decided to explore the neck. An incision was made along the anterior border of the left sternomastoid. The omohyoid muscle was divided and the carotid sheath retracted laterally. The left lobe of the thyroid gland was mobilized and displaced medially. The foreign body could not be felt on palpating the pharyngeal wall. The inferior constrictor muscle was divided along the posterior edge of the inferior cornu of the thyroid cartilage, exposing the mucosa of the pyriform fossa, but the foreign body could not be found. Finally a Michel's clip was attached to the inferior cornu of the thyroid cartilage and the wound was packed with vaseline gauze. Further X-rays were taken on the following day, and these showed that the marker was in close proximity to the foreign body (Fig. 3, *a* and *b*).

A further exploration was undertaken. The mucosa of the pyriform fossa was displaced backwards to expose the posticus muscle, and on separating the fibres of this muscle the foreign body was found and removed. It proved to be a sliver of metal measuring about 2 in. in length (Fig. 4).

The wound was drained for 48 hours; no leakage occurred and the patient was swallowing normally on the fourth post-operative day. Intramuscular penicillin injections were commenced on the day of admission and continued for

(*a*)

(*b*)

FIG. 1.
Lateral and A.P. X-rays of neck showing the position of the foreign body.

FIG. 2.

Lateral X-ray of neck taken after the œsophagoscopy showing that the foreign body has not changed in position. An increase in the width of the post-cricoid soft tissue shadow is apparent.

(*a*)

(*b*)

FIG. 3.

Lateral and A.P. X-rays of neck showing the relation of the Michel clip marker to the foreign body.

five days. On the third day after the operation he developed a deep vein thrombosis of the left calf, but this had settled completely by the tenth day. He had a hoarse voice post-operatively and indirect laryngoscopy revealed a left recurrent laryngeal palsy. The recurrent laryngeal nerve had been identified during the second operation and retracted.

The foreign body appeared to be a sliver of metal from the edge of the pilchard tin, and on further questioning the patient said that he had opened the tin with a pair of metal shears. The foreign body must have entered the left pyriform fossa and come to lie in an antero-posterior horizontal plane. It must

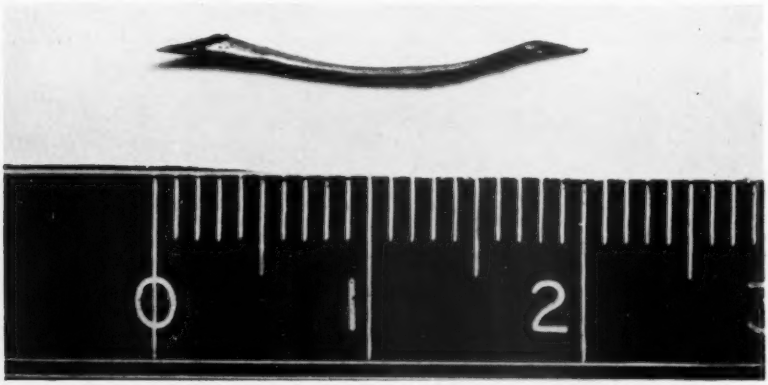

FIG. 4.
The foreign body after removal.

then have been forced through the antero-lateral wall of the fossa, probably by the patient's efforts to expel it, and so have been driven into the substance of the left posticus muscle.

There was no sign of recovery of left vocal cord paralysis three months after the operation, but when he was examined on *August 2nd, 1960*, it was seen that the paralysis had made a complete recovery.

Acknowledgments

I would like to thank Mr. G. H. Bateman for allowing me to publish this case and for his help and guidance in preparing this short paper.

I also wish to thank the X-ray Department at St. Thomas' Hospital for the loan of the X-rays, and the Department of Medical Photography for their help in preparing the photographs.

E.N.T. Department,
St. Thomas' Hospital,
London, S.E.1

HÆMANGIOMA OF THE SUBGLOTTIC SPACE IN AN INFANT AS A CAUSE OF STRIDOR

By GAY, I. and ELIAN, E. (Jerusalem)*

Among the rare causes of stridor in infancy may be cited hæmangioma of the subglottic space. Belonging to the true angiomata they are of a more distinct type than that of the adult, which are situated principally about the glottic aperture. Again, no more than 15 proven cases could be found by us in the literature, while in the adult it is not an uncommon condition. It is possible that many cases may go undiagnosed, and that the condition is more common than the literature would suggest. We would like to add to the limited knowledge gleaned by presenting a further case.

Case Report

A.E., a female infant, was hospitalized at the age of two months because of a stridor and increasing dyspnœa for five days. The infant had a normal delivery following a normal pregnancy. The birth took place at another hospital. Family history was irrelevant.

The physical examination revealed a fairly well developed female infant, weighing 5,500 gm., in obvious respiratory distress, with a marked stridor and with retractions of the suprasternal notch and of the lower intercostals. The voice was clear. A hæmangioma involving the lower lip and some bluish striae in the mental region, probably belonging to the hæmangioma were noted (see Fig. 1). There was evidence of an upper respiratory infection with inflammation of the pharynx and a few rales were found in both lungs.

Intensive antibiotic therapy was instituted, penicillin, streptomycin and chloromycetin, as well as cortisone and water vapours. A direct laryngoscopy done 12 days after admission showed a cushion-like subglottic swelling, somewhat dark in colour. During the course of treatment the dyspnœa disappeared and the stridor diminished. Following a short period of ulceration of the haemangioma of the lower lip, the child was discharged and transferred to our follow-up clinic.

Five days later she was re-admitted because of stridor and dyspnœa with the same clinical picture as on her first admission. At a subsequent laryngoscopy, the same appearance as previously was noted, namely, a darkish cushion-like swelling under the mucosa of the subglottic region, with a normal glottic appearance without signs of laryngitis. This swelling occupied the posterior and left lateral wall.

* From the Departments of E.N.T. and Pædiatrics, Rothschild Hadassah University Hospital, Jerusalem.

Biopsy was not performed because it was considered likely to precipitate bleeding.

Bronchoscopy, which could have shown the lower border of the swelling, was not done as it was considered likely to add to the swelling and precipitate a tracheotomy.

On the basis of the diagnosis the child received X-ray radiation to the

FIG. 1.

subglottic area, a total of some 600 rs. divided into 5 doses. It was interesting to observe the very rapid improvement of the child with the complete disappearance of the stridor following radiation.

There was a follow-up, and the child showed no relapse in a period of 18 months.

Discussion

The literature has been revised and cases presented by Suehs and Herbut (1940); Ferguson (1944); and P. Doermann *et al.* (1958). All the 15 cases presented in the literature have been under the age of one year and the majority (12 cases)

under six months. It is feasible to consider these hæmangiomata as congenital in origin. All presented with dyspnœa and stridor which frequently showed a remittent course. About half of the cases did not show presence of haemangiomata elsewhere and presumably gave more difficulty in diagnosis. In a number of cases also diagnosis was not made until autopsy.

Treatment has been successful following deep X-ray preceded by tracheotomy, radium applied over front of neck, and surgical excision with diathermy in a laryngo-tracheo fissure operation. Two cases have been successfully treated by surgery, one by Rutherford and Sunderland and one by Sharp. In the latter, the infant had a subsequent paresis of the right cord, and the submucosal tumour was removed with sacrifice of overlying tracheal mucosa. Although there was no subsequent stenosis there is no doubt that this is an operation which in infants is fraught with many dangers. In the case presented by Hofmann (1938) treated by laryngo-tracheal fissure, the infant died following wound suppuration and pneumonia.

Sweeter (1921) who was the first to classify infantile and adult types of hæmangioma intubated his patient with subsequent hæmorrhage and death by suffocation.

In Shirley's case cited by Sweeter the hæmangioma was encountered in the tracheal incision and fatal hæmorrhage occurred.

However, New and Clark (1919) successfully treated by intubation combined with radium therapy applied to front of neck with lead screening. They recommend radium placed against tumour by suspension laryngoscopy.

In general, irradiation therapy has been used successfully in five cases. Doermann (1958) recommends a preceding tracheotomy as does Kasabach (1945) to obviate the danger of obstruction from œdema during therapy. There is no doubt about the value of tracheotomy as a life-saving measure, but tracheotomy in the small infant carries its own risk, in addition to the possible risk involved in cutting through a low-lying hæmangioma.

We did not employ tracheotomy and the infant was under close observation in hospital during irradiation therapy. There was no increase of stridor, and presumably no reactionary swelling during deep X-ray therapy.

Summary

A case is presented of subglottic hæmangioma occurring in an infant, and presenting with dyspnœa and stridor. There was an associated hæmangioma of lip and chin area. Treatment was successful following deep X-ray therapy, without a preceding tracheotomy.

Acknowledgments

We wish to thank Dr. M. Feinmesser and Dr. S. Berman of the E.N.T. and Pædiatric Departments of the Rothschild Hadassah University Hospital for their kind permission and advice in the preparation of this case, and to Professor Hochman of the Radium and Tumour Institute, who was responsible for the rœntgentherapy.

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9 Wigram Road,
Wanstead,
London, E.11

CLINICAL NOTE

SINUS TRANSILLUMINATION: AN ARCHAIC SURVIVAL

By M. E. N. SMITH (Swansea)

"The superstition in which we were brought up never loses its power over us, even after we understand it."—LESSING.

THE following extracts are taken from W. G. Porter's *Diseases of the Throat, Nose and Ear* published in 1912, of which "Logan Turner" is the modern version.

"Trans-illumination was introduced by Voltolini and elaborated by Heryng, and is used for the investigation of the condition of the accessory sinuses. The examination has to be carried out in a dark room. The lamp is introduced into the mouth of the patient who then closes his lips. Any denture must be removed first. On making the contact, a semicircular tâche is seen under each lower eyelid if the antra transmit light; the pupils are also slightly illumined, and, if the patient closes his eyes, he has a sensation of light in them. A faint patch of light is also seen on either side of the root of the nose, due to light passing through the ethmoid labyrinth. An antrum may fail to transmit light if it is full of pus, if the walls are thick, or if it contains a solid tumour. The illumination is brighter than usual in cystic conditions, and it may be so in cases of choanal polypus arising from the interior of the antrum."

The following two quotations from the same source refer to the frontal sinus.

"A failure to illuminate conveys no information, as it may be due to the absence of a sinus, or to disease."

"Trans-illumination does not give any assistance in arriving at a diagnosis; a skiagram affords the only conclusive proof of the presence of disease that can be obtained apart from an actual operation."

These extracts exemplify the chaotic basis and fallaciousness of the procedure, namely, that whatever the result it neither proves nor excludes the presence of disease! What diagnostic test could be more inane? Yet for three-quarters of a century it has withstood ridicule, the perfection of finer diagnostic tools, and the tragic lessons to be learnt by those with eyes to see and mind to analyse. The number of lives that have been lost through blind adherence to an archaic anachronism cannot be computed. I believe an important factor in its retention is its "convenience" in private practice. In a private consulting room a trans-illuminator is a possibility while an X-ray apparatus usually is not. It is so much easier to trans-illuminate in the consulting room and act directly upon the result rather than to investigate properly by referring the patient to hospital or a private radiologist for X-ray. How many E.N.T. surgeons would have their own antra punctured without convincing radiological evidence of disease?

This paper has been prompted by the recollections of catastrophe over the years, in the capacity of onlooker let me hasten to add, of those mistakes by which we all obtain our experience. Two sad cases of vivid memory were young women in their early thirties. In one, a right antrum proved dull upon trans-illumination. "Infection" was diagnosed and the patient subjected to a protracted series of antral washouts which produced foul pus upon each occasion and seemed therefore to substantiate the diagnosis. By this means the truth remained obscured for several months until facial pain and swelling led to open exploration of the antrum and the discovery of an extensive, stinking, necrotic carcinoma beyond all hope of radical surgery. Radiotherapy was of no avail.

The other case was distressingly similar in outline but with an additional feature making it even more deplorable. An X-ray had been taken relatively early when the "sinusitis" diagnosed by trans-illumination showed no adequate response to repeated lavage. But the neoplastic erosion of the antral walls was misdiagnosed as "osteomyelitis" and the lavages continued unabated! The eruption of the tumour into the nasal fossa eventually revealed the truth. Such clinical unconsciousness is indeed invincible and in its presence even X-rays are vitiated. In cancer surgery today "Early diagnosis" is the recurring cry. One of its enemies is the trans-illuminator. While we patiently await the triumphant outcome of "Cancer Research" we can help just a little by hastening to reject all impedances to early diagnosis as soon as they are seen for what they are. I offer that as my excuse for this paper.

There is one other, relatively uncommon, condition not so far mentioned, to which the present considerations equally apply, and that is antral rhinolithiasis. This has recently been reviewed by Brain (1960).

I conclude with a grave speculation. In the event of litigation arising out of such a case as I have described, What defence could the surgeon offer to a charge of criminal negligence? Has he given the patient the advantages of modern medical knowledge to which he is entitled? I venture to suggest that he has not and that he has no defence.

Summary

Trans-illumination is out of date. Its continued use without X-ray is a cause of loss of life and probably amounts to criminal negligence with no defence in Law.

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GENERAL NOTES

BRITISH ASSOCIATION OF OTOLARYNGOLOGISTS

Poliomyelitis Inoculation and the Removal of Tonsils and Adenoids

The Council recommends that children should be inoculated against poliomyelitis before the removal of tonsils and adenoids. As a safeguard the ear, nose and throat specialist should ascertain from the parent whether the child has received its second inoculation.

It is appreciated that no direct pressure can be brought to bear on parents if they do not wish to have their child inoculated and they can only be warned of the possible risks.

PROPOSED HEARING AND SPEECH CENTRE AT THE ROYAL NATIONAL THROAT, NOSE AND EAR HOSPITAL

The Trustees of the Nuffield Foundation have offered a grant of £60,000 for the erection of a Hearing and Speech Centre attached to the Royal National Throat, Nose and Ear Hospital and its associated Institute of Laryngology and Otology.

The new Centre will be concerned with the diagnosis, assessment and development of hearing and speech in children suffering from impairment or total loss of these faculties.

The Centre will also be actively engaged in research into the whole problem presented by congenital and acquired deafness, and with the special training in this field of doctors and other professional workers such as psychologists, teachers of the deaf, speech therapists, health visitors and social workers. It is hoped that the Centre will generally be a focal point for all who are concerned in the care of the deaf child.

Building work will commence within the next few months.

ROYAL COLLEGE OF SURGEONS OF ENGLAND

An Arris and Gale lecture with be delivered on February 9th, 1961 by Mr. P. H. Golding-Wood entitled "Autonomic Surgery of the Ear and Nose".

General Notes

VIIème CONGRÈS INTERNATIONAL O.R.L.

LE VIIème Congrès International O.R.L. se tiendra du 23 au 29 juillet 1961 à Paris, à la Nouvelle Faculté de Médecine, Rue des Saints-Pères, sous la présidence de M. le Professeur AUBRY.

Trois très importants rapports seront discutés:

1°) Les acquisitions récentes en pathologie et en thérapeutique de l'otosclérose.
Rapporteurs: MM. les Professeurs SOURDILLE, SERCER and KRMPOTIC, WEBER, ARDOUIN, LARROUDE, FERRERI and SCHAMBAUGH.

2°) Indications et résultats après 5 ans, de la chirurgie et des associations radio-chirurgicales dans le traitement des cancers du larynx et de l'hypopharynx.
Rapporteurs: MM. les Professeurs PIETRANTONI and AGAZZI, ALONSO, CASADESUS, CHEVALIER-JACKSON, LEICHER, LEROUX-ROBERT, ORMEROD.

3°) Rapports entre l'allergie et les infections du nez et des bronches.
Rapporteurs: MM. les Professeurs VAN DISHOECK, CLERICI, HLAVACEK, VOOHORST, MAYER, LASKIEWICZ, TERACOL.

De nombreuses réunions et festivités sont envisagées pour les Congressistes et leurs épouses. A la suite du Congrès, des excursions auront lieu; elles seront proposées dans un avant-programme et réalisées par les soins des WAGON LITS COOK, 22 rue de l'Arcade, Paris, qui sont notre agent officiel habilité pour les transports et réservations d' hôtels.

Pour tous renseignements, écrire au Dr. Henry GUILLON, Secrétaire Général, 6 avenue Mac-Mahon, Paris.

VIIth INTERNATIONAL CONGRESS OF OTO-RHINO-LARYNGOLOGY

THE 7th International Congress of Oto-Rhino-Laryngology will be held in Paris on July 23rd-29th, 1961.

A preliminary circular regarding this Congress has already been sent to all ear, nose and throat specialists in the world. Only those who return the reply form attached to this preliminary communication will receive further details.

Those ear, nose and throat specialists who, owing to an error, have not already received this first communication are asked to write to the Secretary-General if they wish to receive information about the Congress.

Henry Guillon, Secretary-General, VII[e] Congrès International D'Oto-Rhino-Laryngologie, 6 Avenue Mac-Mahon, Paris XVII[e], France.

THE INSTITUTE OF LARYNGOLOGY AND OTOLOGY

330-332, GRAY'S INN ROAD, LONDON, W.C.1.

SHORT COURSES

for

CONSULTANTS AND SENIOR REGISTRARS

7th, 8th and 9th March, 1961—Diagnosis and treatment of the deaf child.
28th and 29th April, 1961.—Practical Aural Surgery Class.

General Notes

NON-MEDICAL COURSES

31st May, 1st and 2nd June, 1961—Hearing and mental assessment of the deaf child—for psychologists.

The number of entries is limited in each course. A detailed syllabus of the above can be obtained on application from the Secretary-Administrator.

SPECIAL LECTURES

Saturday, 4th February, 1961, at 12.00 noon.

Professor Donald R. Griffin—Professor of Zoology, Harvard University, "Echolocation in bats, with special reference to insect catching".

Saturday, 6th May, 1961, at 12.00 noon.

Professor Otto Löwenstein (Birmingham), "The evolution of static and acoustic sense organs".

UNIVERSITÀ DEGLI STUDI DI BOLOGNA
PREMIO INTERNAZIONALE "PIETRO CALICETI"

Announcement of the conditions for the International Prize Pietro Caliceti

The conditions for the "International Prize Pietro Caliceti" are herewith announced. The prize honours the memory of Prof. Pietro Caliceti, deceased distinguished Professor of Clinical otorinolaringojatrica of the University of Bologna.

The prize will be awarded to an Italian or foreign graduate in Medicine and Surgery, who during the four-year period of time (November 1, 1956 to October 31, 1960) has or will publish a scientific work of unquestionable value and of recognized originality in the field of otorinolaringologia.

The prize consists of: (*a*) a gold medal, minted from the original mould used by Mrs. Inez Misley, the widow Caliceti; (*b*) a cash prize of 1.000.000 Lira; (*c*) Diploma testifying to the receipt of the prize itself.

The Committee of Judges will be composed of: (1) The Dean of the Faculty of Medicine and Surgery of the University of Bologna. (2) The Professor of Clinical Otorinolaringologia, of the University of Bologna. (3) The President of the Italian Society of Otorinolaringologia. (4) The Professor of Clinical Surgery of the University of Bologna. (5) The professor of Human Physiology of the University of Bologna.

If in the final judgement of the Committee, none of the works presented by the competitors demonstrates outstanding value and originality, they may decide not to award the prize.

By April 10, 1961 the competitors must send to the "Segreteria dell'Università di Bologna" the following documents: (*a*) an application addressed to the Magnifico Retoret dell' Università degli Studi di Bologna; (*b*) a curriculum vita; (*c*) six copies of each scientific work submitted for the examination for the prize (five of these will go to the members of the Committee of Judges).

In addition, the Committee of judges may give consideration in awarding the prize to informations and data submitted by the above mentioned date by foreign or Italian Universities or by the three societies of Otorinolaringologia: Italian, Latin, International.

TEMPLE UNIVERSITY SCHOOL OF MEDICINE

PHILADELPHIA 40. PA.

A 'Short course in Audiology" is offered several times each year by the Section of Audiology, Department of Otorhinology, Temple University School of Medicine, Philadelphia 40, Pa.

The course will be one week in length and will encompass theoretical and clinical aspects of audition. Hearing testing and interpretation will be stressed. Office procedures, equipment and Industrial Audiometry will be discussed.

Further information may be obtained by writing to Philip E. Rosenberg, Ph.D., Director, Section of Audiology, Temple University Medical Center, 3401 North Broad St., Philadelphia, Pa.

POSTGRADUATE COURSE IN ALLERGY

A continuous course of two weeks duration beginning 8th January, 1961, is being offered by the Departments of Allergy and Applied Immunology of the Temple University Medical Center and the Graduate School of Medicine of the University of Pennsylvania.

For brochure and application forms write to Dr. George Blumstein, c/o Temple Medical Center, Philadelphia 40, Penna.

ROYAL COLLEGE OF SURGEONS
OTOLARYNGOLOGY LECTURES

THE following lectures, arranged jointly by the Royal College of Surgeons of England and the Institute of Laryngology and Otology, will be delivered in the Lecture Hall of the College on Thursday afternoons at 5.30 p.m.

1961

February 2nd. PROFESSOR VICTOR LAMBERT. "Malignant Melanoma of the Nose."

March 2nd. PROFESSOR L. P. GARROD. "Principles of Antibiotic Therapy in Surgery."

May 4th. DR. R. BODLEY SCOTT. "Blood diseases in relation to Otolaryngology."

General Notes

ROYAL NATIONAL THROAT, NOSE AND EAR HOSPITAL

The Trustees of the Nuffield Foundation have increased from £60,000 to £100,000 their grant for the erection of the Nuffield Hearing and Speech Centre attached to The Royal National Throat, Nose and Ear Hospital in Gray's Inn Road.

It is expected that building work will commence early in 1961 and it is hoped that the new centre, which is unique in conception and design, will be a rallying point for all concerned in the welfare of deaf children.

THE ROYAL NATIONAL THROAT, NOSE AND EAR HOSPITAL

KING EDWARD'S Hospital Fund for London has made a grant of £20,000 for the purchase and adaptation of No. 6 Castlebar Hill, Ealing for use as a hostel for deaf children who are under treatment and training by the Audiology Unit of the Royal National Throat, Nose and Ear Hospital.

The new hostel (which adjoins the existing hostel for mothers and very young deaf children) will accommodate 20 children of varying ages and will enable longer and more intensive periods of training to be given than is at present possible. There will be facilities for mothers, and occasionally fathers, to stay at the hostel with the children from time to time.

The work of adaptation is to be started early in the New Year in the hope that the hostel will be ready for opening in April or May, 1961.

UNIVERSITY OF EDINBURGH FACULTY OF MEDICINE

THE LLEWELLYN BEVAN PRIZE

This Prize, of the value of about £80, will be open for award in 1961 to a Graduate in Medicine of the University of Edinburgh for the best and approved Essay on the Pathology and Treatment of Ménière's or other Disease of the Internal Ear.

Essays in competition for the Prize must be submitted not later than October 31st, 1961, to the Dean of the Faculty of Medicine, University, Teviot Place, Edinburgh, 8.

General Notes

EXAMINING BOARD IN ENGLAND BY THE ROYAL COLLEGE OF PHYSICIANS OF LONDON AND THE ROYAL COLLEGE OF SURGEONS OF ENGLAND

Diploma in Laryngology and Otology

The Secretary reports to the two Royal Colleges that at the First Part of the Examination held in December, 1960, 54 Candidates presented themselves, of whom 29 passed; and that at the Second Part of the Examination held in December, 1960, 40 Candidates presented themselves of whom 25 passed.

The following 24 Candidates, having complied with the regulations of the Board, are eligible to receive the Diploma, viz.:

Atkins, John	9 Hawthorn Park, Handsworth Wood, Birmingham, 20.
Boyle, Michael James	Royal National Throat, Nose and Ear Hospital, Gray's Inn Road, W.C.1.
Edwards, William Glyndwr	12A Kirkdale, S.E.26.
Ferris, Keith Parham	37 The Priory, S.E.3.
Gill, Balraj Singh	Ophthalmic Hospital, Maidstone, Kent.
Griffiths, Horatio Jonathan	60 Copthorne Park, Shrewsbury, Shropshire.
Gyaw, Christopher Pee	19 Build. 6, Lanthit Yeiktha, Rangoon, Burma.
Hasnain, Syed Wahaj-ul	35A Preston Road, Westcliff, Essex.
Hayes, Arthur Vivian	11 Sandhurst Road, Vincent, East London, Cape Province, S. Africa.
Hilmi, Suhail Rasheed	66 Greencroft Gardens, N.W.6.
Hla, Maung San	13 Panchan Street, Sanchaung, Rangoon, Burma.
Jackson, James McGillivray	11 Claremont Place, Newcastle-on-Tyne.
Jayarajah, Samuel Charles	St. Peter's Hospital, Chertsey, Surrey.
Jog, Mohan Ganesh	Royal Hospital, Wolverhampton.
Landeg, Mervyn Russell	83 Sandown Road, Port Talbot, Glam.
Mathen, Kayyalakakathu Mathen	St. Mary Abbotts Hospital, W.8.
Morris, Thomas Michael Owen	Brambletye, Poundhill, Worth, Sussex.
Owen, David Roy	The Vicarage Flat, Wilstead, Bedford.
Ramachandra, Vaitilingam	240/4 Torrington Avenue, Colombo, 7.
Thar Hlaing	32 Bolton Gardens, S.W.5.
Tucker, William Nichol	c/o Bank of New Zealand, 1 Queen Victoria Street, E.C.4.
Wijeyaratne, Donald Stanley	Seethala Agara Estate, Dunagaha, Ceylon.
Wilson, Ian Irvine	16 Glenhill Close, Lichfield Grove, N.3.
Yorston, Robert Allan	Templehall, Longforgan, by Dundee, Angus.